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THE PROBLEMS OF URETERAL TRANSPLANTATION

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URETERAL transplantation may be indicated when lesions of, or involving the urinary tract cannot be remedied to allow function within a relatively normal range. Interference with ureteral clearance will inevitably lead to renal deterioration. Failure of the bladder to act as a reservoir of convenience can render life completely intolerable, as can lack of control due to urethral pathology. In such cases diversion of the urine may be the only means of preserving renal function and social status. However, the attempted cure or palliation of neoplastic disease by vesical ablation or exclusion, provides the commonest indication for the measure. The diversion may be intestinal or non-intestinal and for practical purposes, drainage to the lower bowel or to the skin represent current technique. Either operation should only be performed in the absence of any reasonable alternative treatment. For completeness two other procedures for the disposition of the ureter must be mentioned, namely, re-implantation to the bladder and trans-uretero-ureteral anastomosis.

Re-implantation may be possible following injury in the juxta-vesical region. Most commonly, this results from surgical accident and is rarely recognized at the time. Immediate repair or deligation is the ideal but in most instances re-implantation at a later date has to be attempted. The operation may also be indicated when removal of a diverticulum involves the ureter or when the ureteral termination is ectopic and extra-sphincteric. It is important to implant the ureter in the base or fixed part of the bladder. Patton (1939) recommended and reported having used the intra-mural bed. His patient had excellent renal function eight and a half years later. This trans-meatal technique is only possible when sufficient ureter is available for its performance without any tension. Ockerblad (1947) reported the use of a tubed tongue of bladder to replace a ureteral loss of about ten centimetres. The operation was performed in 1936 and eleven years later the drainage and function of the associated kidney were within normal limits. Millin (1950) was able to bridge a gap of about the same extent on each side by this method which was originally suggested by Boari of Italy in 1894.

Trans-uretero-ureteral anastomosis was first carried out by Higgins in 1934. The implantation of the damaged ureter in its healthy fellow is performed retro-peritoneally and raises the question as to whether interference with the single normal ureter is justified. The success achieved in a few cases may warrant occasional use of this method of conserving renal function in the event of damage to one lower ureter.

Cutaneous Ureterostomy

Diversion of the urine by cutaneous ureterostomy is used less frequently than drainage to the bowel but is employed when rectal control is deficient or when ureteral pathology, such as marked dilatation or shortness, preclude lower bowel implanta-

tion. This method possesses the disadvantage, that the uncontrolled flow of urine imposes a problem of collection and disposal, together with a toilet of cleansing and sterilization. However, drainage to the skin surface is more simple, less hazardous and associated with less distressing morbidity. Stenosis of the terminal ureter is easily dilated, infection of the kidney less fulminating and serious whilst there is no alteration of blood chemistry as so often occurs in uretero-colic anastomosis.

Mathé (1944) reported survivals of sixteen and twenty-three years following cutaneous ureterostomy. A male patient of mine recently died aged fifty-five years, having lived fourteen years with his solitary left kidney draining to the skin by ureterostomy.

The point of emergence of the ureter should be above the normal belt line and sufficiently anterior to allow of convenient supervision by the patient. The course to the skin should be direct and angulation avoided. Fish and Stevenson (1948) suggested ureterostomy through a cutaneous pedicle graft to facilitate the collection of urine.

INTESTINAL TRANSPLANTATION

Transplantation to the bowel has been performed more often than to any other organ and most frequently to the intact sigmoid. Chaput of France, a pioneer in developing the procedure, wrote in 1894, "The operation is one of necessity, not of choice and is only done when nothing else can be offered to preserve kidney function." Despite improvements in surgical technique and the assistance of antibiotic cover, the measure itself still carries appreciable mortality, imposes an altered habit and often creates grave physiological imbalance. It is still emphatically an operation of necessity.

The risk of ascending infection from the faecal stream has long been appreciated and numerous attempts have been made to provide a less septic reservoir than the functioning bowel. Partially, or completely excluded segments of small and large intestine have been used to replace the extirpated bladder, since the first attempt of Tizzoni and Foggi (1888). Bricker (1950) isolated the terminal few inches of the ileum in human patients, after pelvic exenteration and transplanted the ureters to this intra-abdominal reservoir. Boyce and Vest (1952), in the treatment of exstrophy, recommend the creation of a permanent colostomy and the use of the excluded lower third of the sigmoid as a urinary reservoir. Without disturbance of the ureteral orifice, they have anastomosed the bladder to the rectum, virtually adding it as a diverticulum to that organ. Although such artificial bladders possess many deficiencies, this work generally is impressive and if developed may remove some of the important disabilities of uretero-colic-anastomosis.

Despite physiological shortcomings, drainage to the intact lower bowel and its use as a urinary reservoir is an established surgical measure. John Simon achieved diversion of urine to the rectum in 1851 by suture fistularization and Smith, also of England, directly anastomosed ureter to bowel in 1878. The principle introduced by Simon has since been used by Coffey (1930) in his third technique and by Higgins (1933) in transplanting the intact ureter. The mucosa to mucosa, end to side technique of Cordonnier (1949) and Nesbit (1949) embodies the principle introduced by Smith. Bardenheur (1887) developed the muscularizing principle by forming a muscular canal around the implanted ureter in a manner similar to the gastrostomy of Witzel. Maydl (1892) transplanted the trigone and Bergenhem (1894) the intact ureteral orifices to the rectum.

In 1909 Coffey suggested the technique of sub-mucosal transplantation, the ureter entering the bowel obliquely in imitation of its insertion in the bladder. Within a few years he and Mayo established the method clinically and, with minor variations, it has proved the most generally used of all types of transplant. Cabot (1921) suggested extra-peritoneal implantation and Leadbetter (1950) reported having combined mucosa to mucosa union with the use of a sub-mucosal tunnel.

Hinman and Weyrauch (1936) surveyed uretero-intestinal anastomosis in a comprehensive presentation from which I have drawn the historical information. They

recognized eleven principles as having been used but, in 1939, Hinman accepted four as having been most generally applied. They are:—

1. Implantation of the intact orifice.
2. Use of the muscular canal.
3. Oblique sub-mucosal implantation.
4. Implantation of the intact ureter.

To these may be added the end to side mucosa to mucosa transplantation, which has enjoyed popular favour since re-introduced by Cordonnier and Nesbit.

It is of interest that A. A. Lendon, of Adelaide, at the Inter-Colonial Medical Congress held in Hobart in 1902, reported having transplanted the ureters of 4 patients. The first operation was performed on 12th May, 1899. Recently, by the kindness of Mr. Edye, I was able to examine a male aged twenty-eight years, born with complete epispadias and exstrophy. Twenty-two years ago Sir Alexander McCormack performed Bergenhem's operation and transplanted the intact ureteral orifices to the rectum. The patient is the father of two children, is very well and leads an active sporting life. The excretion urogram made recently shows bilateral renal function to be excellent with a minor degree of ureteral and pelvic dilatation on the right side.

Hinman (1939) reviewed several hundred cases and concluded that the results were slightly superior using the sub-mucosal principle of Coffey but not enough to prove that the problem had been solved by any method or technique. Obviously, the mucosa to mucosa method and Leadbetter's modification have not been used long enough for assessment.

The operation should not be performed with impairment of rectal control while advanced age, general debility and renal insufficiency are relative contra-indications.

In general, any ureteral lesion which does not allow free delivery of urine to the bladder may necessitate transplantation to the bowel. Often damage results from surgical accident and, in this connection, I believe the attitude of the medical profession should be changed. Ligation or division of a ureter during a difficult pelvic operation has occurred sufficiently often, despite all reasonable precautions by fully qualified surgeons to be accepted as a legitimate operative complication. I am sure the medico-legal suits in this connection will be less, if, when damage to the ureter occurs and the clinical course altered as a result, the patient is informed.

Bladder pathology furnishes the commonest indication for diversion of urine. Of the congenital conditions, exstrophy, complete epispadias and other anomalies in which sphincteric control is lacking, warrant the operation as they deny the bladder its function as a controlled reservoir. Exstrophy, which occurs once in about 40,000 births provided for many years the main stimulus to the development of the procedure. Apart from the extreme social handicap of this condition, inevitable renal damage from infection makes surgical treatment imperative. Untreated, more than half the children die before the age of ten years and few reach the age of twenty years. Grey Turner (1943) reported survivals of 9 patients of periods from sixteen to thirty years. All had been born with defects resulting in complete incontinence. Unfortunately, the report did not state the total number of patients treated. Higgins (1943) advised early operation and stressed the advantages of its performance during the first twelve months of life. His conclusions would appear to be generally accepted. I prefer in the very young, to use the extra-peritoneal method of Cabot and to operate on the ureters separately, allowing an interval of two or three weeks between each transplantation.

Acquired pathology for which uretero-colic anastomosis may be necessary, is simple or neoplastic. In the former, irreparable fistulae and extensive injuries to the bladder or urethra, to render control impossible, provide clear cut indications. Extreme bladder contracture, resulting from chronic inflammation, whether it be tuberculous or not may justify vesical exclusion. In addition, radium or X-ray therapy to other pelvic organs may seriously damage the bladder and deprive it of any useful function.

Transplantation of the ureters is performed more often in the treatment of bladder growths than for all other conditions combined. The approach may be palliative when

complete removal of the neoplasm is known to be impossible or in association with cystectomy when cure by radical surgery is attempted. In the former, by diverting the urine and removing any physiological stimulus to the bladder considerable relief may be afforded. Further, marked regression and even disappearance of a cancer has been reported as having occurred after vesical exclusion. Goldberg (1950) was unable to find any evidence of a previously observed infiltrating growth some weeks after transplantation of both ureters. Mortensen (1952) reported dramatic regression with disappearance of symptoms for about a year in two cases. After that period the growths pursued their malign course.

Opinion is divided as to when radical surgery is warranted in the treatment of bladder neoplasms. In the obviously incurable case diversion of urine may or may not give relief and the procedure, even as a last resort must be accepted as being quite speculative. I believe radical surgery, by which I mean ureteral transplantation and total cystectomy, to be illogical and unjustified in the treatment of most bladder growths. Hinman (1939) said, "If a cancer can be cured by less radical measures the procedure is inapt and if a cancer cannot be all removed the procedure is useless." This conception is basic but does not appear to have been appreciated generally. In consequence, a review of urological literature is not helpful in evaluating treatment as I have gained the impression that transplantation and cystectomy are performed very often in the presence of incurable malignancy and that operative technique is frequently regarded as being more important than prognosis.

Jewett (1944) stressed the relationship of the depth of infiltration of bladder growths to potential cure. In an exhaustive pathological and clinical study of 206 cases, he found the depth of invasion to conform generally to a prognostic pattern, whereas cellular differentiation was only helpful in about 50 per cent. The patients were divided into two groups. In one the growths had infiltrated less than half way through the bladder musculature and in the other more than half way or beyond the bladder wall. In each group complete surgical extirpation of the neoplasm was attempted but with very different results. In the superficially infiltrating group, the five year survival rate was 74 per cent. and only 10 per cent. of the patients died or, at the time of the report, were living with malignant disease. In the other group, three per cent. survived five years and malignant disease had killed or was still present in 85 per cent. of patients.

Jewett suggested that, to the trained observer, bi-manual examination under anaesthesia will afford sufficient evidence to distinguish between the superficially and deeply infiltrating growths. Virtually, the distinction is as to whether the growth is curable or incurable. If palpable as a stony hard or even firm rubbery mass, Jewett considers the growth as deeply infiltrating. Additional prognostic evidence may be gained by radical loop resection of a growth and estimation of depth of invasion by examination of the fragments. This approach may not be ultimate but it is constructive. Certainly, pre-operative assessment of curability must replace the accepted standards of operability. These latter often mean little more than personal ability to transplant ureters and remove a bladder which is not entirely frozen by growth.

The inference that cure is impossible and radical surgery unjustified in deeply invasive growths is supported by many observers. Colston and Leadbetter (1936) and Jewett (1950) showed remote blood borne metastases to result in growths of the bladder although most often the spread is by lymphatics. Anatomical studies support a spreading belief that only the earliest infiltrating growths of the bladder are curable by surgery, whether it be radical or local. The outlook in non-infiltrating growths is better. Parker (1936), in a study of lymphatic drainage of the female bladder and bladder neck demonstrated lymph channels and glands associated with haemorrhoidal and obturator vessels together with a lymph flow to gluteal and ischial regions, suggesting that even local pelvic spread is rarely circumscribed enough to be curable by surgery. Powell (1944) showed the sub-mucosal and superficial muscle layers to possess scanty lymphatic channels, whereas the deeper musculature and the peri-vesical areas are liberally supplied and with much larger

systems. Batson (1940) stressed the role of the vertebral veins in early neoplastic spread to pelvic bones, vertebrae, ribs and skull from primary growths in the pelvis. Although radical surgery to the extent of pelvic exenteration is practised, there has of late been a distinct trend towards conservatism. Milner (1946) reported having treated 245 patients with bladder growths during the preceding five years. He performed trans-urethral excision and fulguration in 93 per cent. Reynolds, Shulte and Hammer (1949) stressed the value of trans-urethral management by radical excision. In a recent personal communication they again emphasized excision rather than electro-dessication or fulguration.

O'Conor (1952) said, "I have been through three distinct phases of enthusiasm followed by disappointment for so called radical treatment in carcinoma of the bladder." He expressed the opinion that the procedure did not lengthen the patient's life but accepted ureteral transplantation as a valuable palliative measure.

Jewett (1952) summed up the present position by saying, "In general, the outlook depends largely on whether the growth is confined to the bladder or has spread beyond it. At the present time a reasonably good prognosis can be expected only following the complete eradication of the tumour which has infiltrated less than half way through the muscularis and the control of future occurrences identified by cystoscopy at regular intervals. Radical cystectomy and pelvic exenteration have not been used for sufficiently long periods for any conclusions to be reached in regard to long term survivals. At the moment all that can be afforded patients with a deeply infiltrating tumour is a certain amount of temporary palliation or an extensive pelvic dissection of unknown efficacy."

As neoplastic disease of the bladder provides the commonest indication for transplantation of the ureters, with or without cystectomy, I have devoted rather disproportionate space to this clinical entity. I would stress again the need for an altered approach, the deciding factor to be curability rather than operability. The operation is a serious one. Faecal or urinary fistulae and peritonitis are immediate complications; some mortality cannot be avoided and, in the majority of cases, a remote but possibly lethal morbidity must be accepted. This is borne out by the figures of Colby and Kerr (1951) who assessed the types of treatment of new growths of the bladder. All 16 patients with non-invasive growths, treated by segmental resection or local destruction, survived five years and were free of growth. Of 12 patients with non-invasive growths treated by cystectomy and transplantation, only 2 survived five years and none appear to have died of cancer. For some time I have avoided ureteral transplantation and cystectomy for vesical growths as quite independently of other observers, I had reached the conclusion that equal results could be achieved by less radical measures. The advantage of preserving bladder function does not need to be stressed. I believe there are three indications for ureteral transplantation in the treatment of malignant disease of the bladder. These are:—

1. As a palliative measure to avoid painful spasm associated with filling and emptying of the organ;
2. In association with cystectomy for multiple superficial lesions which cannot be controlled by diathermy;
3. In the cases where the complete removal or destruction of the growth, and this applies to other pelvic growths, results in loss of bladder function.

OPERATIVE TECHNIQUE

Hinman and Weyrauch classified ureteral transplantation according to eleven principles but various modifications have resulted in possibly one hundred different techniques. Certain general principles of surgery must be followed with any method. The sigmoid is the most commonly used site for implantation and although the procedure is often attended by disturbance of electrolyte balance transplantation to the caecum has resulted in more serious upset. Riches (1951) reported 5 deaths following implantation in the caecum.

Recently (1952), Butler and Miller investigated the problem of ureteral reflux. In view of the normal and constant intra-colonic pressure, they questioned the permanence of the competency of any valve created at

operation and advised that the implant should be in the mid descending colon, above the level of accumulated urine.

The lower bowel should be investigated by barium enema and X-ray, if the extra-peritoneal operation of Cabot is planned as a short mesosigmoid may prevent its performance on the right side. Preparation includes the usual supportive measures but with a four or five day toilet of the bowel in addition. Initial catharsis may be necessary, while a low residue and low protein diet with daily lower bowel lavage are established routine. Bowel sterilization should be attempted. A dosage of .5 gm. of streptomycin by mouth 4 times a day is satisfactory, but recently Dearing and Heilman (1950) have recommended a similar dosage of aureomycin. Post-operatively continuous rectal drainage should be effected for five or six days and Wangenstein's suction may be necessary to limit upper tract distention.

At operation, mobility of the recto-sigmoid must be assured so that the bowel may be brought to the course of the ureter and not *vice versa*. The ureter must be accorded every respect. It should be mobilized minimally and 4 to 5 cm. should suffice. Trauma must be avoided and clamps should never be put on the proximal segments. The submucosal trough must be quite adequate in all variations of Coffey's first method and haemostasis in its preparation absolute. Finally, after completing the implant, the ureter must be restored to its normal position and the bowel fixed by liberal suturing to the parietal peritoneum.

Despite all precautions, obstruction may result, the two dangerous points being in the tunnel, by constriction, or at the termination of the ureter by stenosis. Humiston (1951) reported a practical approach to this problem. Post-operative progress was unsatisfactory in a male aged forty-five years after transplantation of both ureters. Renal function was diminishing and bilateral hydronephrosis was developing. Eleven months after the transplantation the abdomen was opened and each anastomosis explored from the bowel aspect through a colotomy. Bilateral stenosis was relieved by intra-colonic plastic operations with dramatic benefit.

Cordonnier and Nesbit, in discarding the tunnel and apparently sacrificing the renal protection it afforded, sought by an end to side anastomosis to prevent obstruction at the two critical points. The operation was also shown to be practicable with markedly dilated ureters. There are, however, major deficiencies in the procedure and in this respect Wisenand and Moore (1951) made some pertinent observations. In a female aged forty-seven years, a large fungating growth caused ureteral obstruction which necessitated bilateral nephrostomy. This was followed by cystectomy and bilateral ureterosigmoidostomy using the end to side technique. Within a few days there was free drainage of faeces and gas from the nephrostomy tubes. After clamping the tubes, the expulsive pressure of gas and faeces reached 40 to 50 mm. of mercury, approximately the figure at which the defaecation reflux occurs. The normal resting intra-colonic pressure is about 20 mm. of mercury, while straining creates a pressure varying from 100 to 200 mm. of mercury. The surgeons said, "We believe that subsequent observations will demonstrate the deleterious effect of free transmission of intra-colonic pressures to the renal pelves."

Leadbetter (1951) reported a small series of cases in which he had combined the tunnel principle with the mucosa to mucosa type of junction. He aimed to avoid terminal stenosis of the ureter but, at the same time by the oblique insertion, to provide renal protection against free intestinal reflux.

BIOCHEMICAL CHANGES FOLLOWING TRANSPLANTATION

When both ureters are transplanted to the bowel, disturbance of the electrolyte balance may occur. Ferris and Odel (1950) found some degree of hyperchloraemia in 79 per cent. of 141 patients, and about the same proportion showed evidence of acidosis with a lowered carbon dioxide combining power. The symptoms are weakness, anorexia, nausea, vomiting and a liability to early fatigue. A salt taste occurs with increased thirst and diarrhoea is not uncommon. Ferris and Odel suggested that a selective

reabsorption of the chloride from the bowel, associated with a relative renal insufficiency, resulted in chloride retention. Impairment of renal function does not appear to be necessary for the syndrome, but when present aggravates the condition.

Boyce and Vest (1952) suggested that the chloride reabsorption is largely in the form of ammonium salts. Korenberg (1951) considered the electrolyte changes as being due primarily to sodium chloride reabsorption. Chloride reabsorption may initiate cellular dehydration with increased potassium loss from the cells and excretion in the urine. When rehydration takes place the potassium loss is made good by sodium.

Wilkinson (1952) pointed out that acidosis has not been recorded in a patient with an incontinent rectal sphincter and has suggested that retention of urine in the colon causes increased colonic secretion. Together with dehydration the loss of alkaline secretion results in potassium and bicarbonate depletion. The dehydration in itself leads to renal retention of sodium and mobilization of intracellular water with potassium loss.

The mechanism is imperfectly understood. Selective bowel reabsorption of urinary products may occur or the imbalance may be initiated as suggested by Wilkinson by potassium and bicarbonate loss resulting from increased colonic secretion. The nett result is a syndrome which adds significant morbidity to the operation of ureterosigmoidostomy.

The following measures are recommended in treatment:

(a) Careful and regular emptying of the bowel.

(b) Alkaline ash diet, to furnish base requirements and reduce the excretion or urea and ammonium.

(c) Low chloride intake.

(d) Administration of alkali. In this respect Wilkinson has recommended thirty grammes each of sodium bicarbonate and potassium citrate 3 times a day for short periods of one week in every four. The dose administered can be varied as required.

SUMMARY

1. Ureteral transplantation, either to the skin or lower bowel may be indicated to relieve distressing, dysfunction of the urinary tract or in association with cystectomy, when total removal of a neoplasm is attempted.

2. Neither operation should be performed when a reasonable alternative is available, as at the best, intolerable existence may be rendered tolerable.

3. Transplantation to the bowel has a definite mortality and often creates grave physiological imbalances.

4. The latter operation is justified only within narrow limits in the treatment of neoplastic disease. Curability should replace operability as a therapeutic indication.

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AN ECTOPIC URETER IN A CHILD

REPORT OF A CASE

By Douglas Cohen

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THE ectopic ureter is an uncommon abnormality, only some 300 cases having been reported. The embryology, diagnosis and treatment has been well covered by Higgins and Alldred (1951), who reported a series of four cases, all females.

Case Report

W.K., a female, aged 10 years, was first seen in May, 1952, and was admitted to hospital the same month.

She had been constantly wet by day and night since birth although her mother thought she was worse during the daytime. She passed urine normally also and had normal bladder sensation. She had been treated previously for "enuresis" with drugs without improvement.

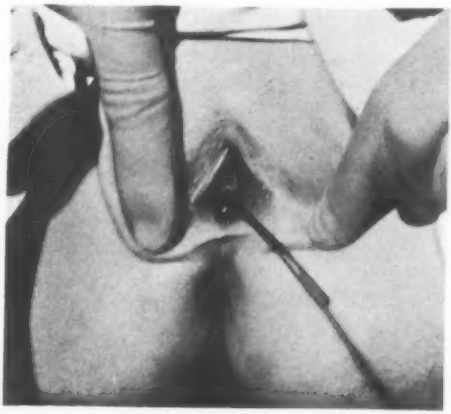

FIG. I. A probe has been inserted into the accessory orifice which is situated just below and to the right of the urethral orifice.

No abnormality was detected on general physical examination except that there was some redness and irritation around the vaginal introitus and urine continually welled up from the vagina and ran down over the perineum. Examination under anaesthesia revealed an ectopic orifice which could be seen just below and to the right of the urethral opening (Fig. I). This opening admitted a probe only with considerable difficulty and eventually a fine ureteric catheter, stiffened with tonsil wire, was manipulated up the ectopic ureter. There appeared to be a point of obstruction about one inch from the surface because, once past this point, the catheter passed onwards without difficulty. Sodium iodide (40 ml.) was injected into the ectopic ureter and radiographs were taken (Fig. II). At the same time cystoscopy and retrograde pyelography were performed (Fig. III). Two normal ureteric orifices were present.

On admission, 0.4 per cent. indigo-carmine was run into the bladder by catheter, a vulval pad was applied and the patient instructed to abstain from micturition. At the end of one hour the pad was soaked with colourless urine, proving quite conclusively that the bladder was not the source of the efflux.

Intravenous pyelography disclosed a partial duplication of the left ureter, the two portions appearing to join about the level of the third lumbar vertebra.

There was also a faint dye shadow above the upper pole of the right kidney in the 30 minute film, but the concentration of the dye by the abnormal renal segment was too poor to outline the ectopic ureter (Fig. IV).

Microscopic examination and culture of the urine collected from the ectopic ureter showed a heavy growth of *Escherichia coli* and numerous pus cells. Examination of the bladder urine revealed no abnormality.

Operation. Exposure of the kidney by the usual loin approach demonstrated an upper and smaller segment clearly demarcated from the remainder of the kidney by a cleft (Fig. V). This segment received its blood supply from a separate leash of vessels arising from the renal vessels and when these were tied delineation of the abnormal segment became even more obvious because of the ensuing congestion in this segment. The dilated ectopic ureter arose from this segment which was now dissected from the remainder of the kidney and the ureter was then followed downwards as far as possible. An oblique incision was made in the right iliac fossa and the normal and ectopic ureters

were again identified. The abnormal renal segment and ectopic ureter were now passed through from the upper to the lower incision and dissected free well down into the pelvis where the ureter began to narrow and, at this point, about three inches below the external iliac vessels the ectopic ureter was transected. Convalescence was uneventful and the child was dry from the time of operation, and has since had no further trouble.

resistant to infection but there will usually be an associated incontinence which will persist after the infection is controlled. These symptoms only occur in females, who comprise the great majority of cases. In the male, if the condition causes any symptoms at all, the story will be of recurrent urinary infections, for incontinence is never a feature.

FIG. II. Ureterogram. Forty ml. of sodium iodide have been injected into the ectopic orifice, outlining a dilated and tortuous ureter. Retrograde pyelography has also been performed.

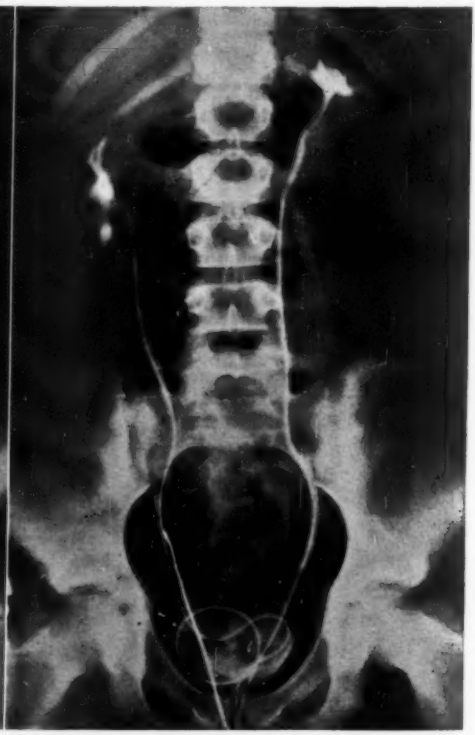

FIG. III. Retrograde pyelogram. Normal outline on the right side. Only the upper calyx has been filled on the left side. A probe has been inserted in the ectopic ureter.

CONCLUSIONS

This uncommon condition should be borne in mind in every case which presents with "enuresis," otherwise it may very easily be missed. In most cases the typical story of "vertical incontinence" may be elicited since the effect of gravity will increase the loss of urine which is pooled in the dilated ectopic ureter. In some cases the patient will present primarily as a recurrent pyelitis which is

In arriving at a diagnosis the most helpful features are a careful inspection of the vulva and urethral orifice. In a sensitive child this may demand examination under an anaesthetic. In most cases the ectopic orifice will be seen adjacent to, or just within, the urethral opening and efflux of urine can be seen. The instillation of indigocarmine into the bladder and the subsequent soaking of a vulval pad with colourless

FIG. IV. Intravenous pyelogram. A faint dye shadow is visible at the upper pole of the right kidney. There is a partial duplication of the left pelvis and ureter.

urine is conclusive evidence that the urine is derived from a source other than the bladder.

If a ureteric catheter can be introduced into the ectopic orifice and a ureterogram taken after the introduction of sodium iodide this affords the most obvious and dramatic proof of the diagnosis. However, much time and patience may be required to effect this. It was found helpful in this case to stiffen the ureteric catheter with tonsil wire prior to its introduction.

Retrograde and intravenous pyelography should always be performed but are of variable help. It is necessary to ascertain how many ureteric orifices are present in the bladder and to be assured of a normal functioning kidney, at least on the opposite side. Usually the abnormal renal segment is not able to concentrate the dye sufficiently to throw a definite shadow, but deformity of the calyceal system or an abnormally low position of the kidney on the affected side may be demonstrated.

Removal of the abnormal segment of renal tissue together with the ectopic ureter, which should be transected as low down as possible, is the procedure of choice, and results in a dramatic cure.

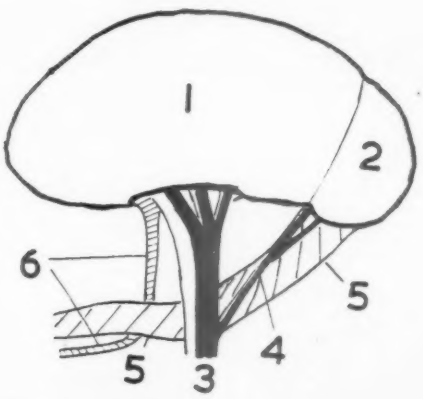

FIG. V. Right kidney exposed at operation. Explanatory diagram shows: 1. Kidney. 2. Abnormal renal segment. 3. Renal vessels. 4. Leash of vessels to abnormal segment. 5. Ectopic ureter. 6. Normal ureter.

FIG. VI. Operation specimen showing excised segment of renal tissue and ectopic ureter.

ACKNOWLEDGEMENT

My thanks are due to Mr. T. Y. Nelson for his advice and assistance with this case.

SUMMARY

1. A case of ectopic right ureter in a female child is described.
2. The features which are of most help in making a diagnosis and the method of treatment are discussed.

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OSTEOCLASTOMA OF THE PETROUS TEMPORAL BONE

REPORT OF A CASE BENEFITED BY DEEP X-RAY THERAPY

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OSTEOCLASTOMA of the petrous temporal bone is a very rare tumour, and the following case is therefore of interest, not only on this account, but also because of the successful outcome of treatment.

Case Report

History

A Syrian girl, aged 8 years, had been well until February, 1951, when she became listless, and vomited several times. A few days later she noticed double vision on looking towards the left side, and about the same time her mother noted that the left side of her face was paralyzed. Shortly afterwards she was admitted to the American University Hospital, Lebanon, under Professor Hassan Idriss. She then showed complete paralysis of the VIth and VIIth nerves on her left side, and also partial deafness of conduction type in her left ear with a slightly bulging eardrum. Special investigations, including blood picture, C.S.F. examination, E.S.R. and W.R. were normal. X-rays of the mastoid region, however, showed diffuse haziness of the left mastoid bone with destruction of the petrous apex. Gradenigo's syndrome was suspected and treatment with antibiotics commenced, but no improvement occurred. Some days later a left myringotomy was performed, followed after seventeen days by a simple mastoidectomy at another hospital. The findings at each operation were indefinite. Her post-operative convalescence was straightforward. However, further radiographs six weeks later showed increased destruction of the petrous apex, and she was referred to Sir Charles Symonds in this country who, in turn, referred her to this Unit.

Examination

When admitted on 17th May, 1951, she looked a robust, healthy girl. We confirmed the left VIIth nerve palsy, including absence of tear secretion and of taste sensibility on the left side; severe deafness of her left ear with absent caloric responses on her left side; a partial left VIth nerve palsy, abduction of her left eye being limited to 20 degrees (Fig. I). Tactile discrimination was slightly impaired over the lower third of her face. X-rays of the skull (Fig. II) revealed complete destruction of the apex of the petrous bone. The erosion extended posteriorly to the carotid canal and antero-laterally to the *foramen ovale* and adjacent parts of the greater wing of the sphenoid. The basi-sphenoid and basi-occiput were intact. A faint line of calcification could be seen intracranially about 1.5 cm. above the petrous bone. These details were clearly seen in the basal projection and Stenver's projection, but were scarcely apparent in the routine P-A, lateral, and Towne's projections.

FIG. I. Pre-operative photograph showing left VIth and VIIth nerve palsies.

Operative Findings

On 23rd May, a sub-temporal craniotomy was performed by Mr. Murray Falconer. A dark red fleshy tumour 5.5 cm. wide and 2.5 cm. high was found occupying the superior surface of the petrous bone and elevating the *dura mater* of the middle fossa. At its posterior margin there was a small amount of new bone formation. When incised, the tumour was seen to be laminated, its layers being separated by a thin film of old blood, and on aspiration its interior yielded about 10 cc. of old blood. The extent of the tumour was such that complete removal was impossible. Several pieces of tumour and adjacent bone were taken for biopsy and the craniotomy then closed.

Histology (Fig. IV)

Dr. S. de Navasquez reported that the sections showed the typical appearances of an osteoclastoma. Multinucleated giant cells were abundant and the stroma, which was highly vascular, consisted for the most part of spindle cells, with some areas of more darkly stained round cells. The surrounding bone showed well marked fibrous replacement with many small cysts. All gradations of this process could be traced from the edge of the tumour out to normal bone.

When discharged from hospital four weeks after operation, her signs were unchanged except that she was now completely deaf in her left ear. On reporting two months later her facial paralysis and deafness were still unchanged. However, Vth nerve sensibility was now completely normal and the VIth nerve palsy had improved. Fresh radiographs showed a striking change, for the defect in the petrous bone was now occupied by a calcified mass projecting like a dome into the middle cranial fossa. In view of this encouraging improvement a further ten deep X-ray treatments were

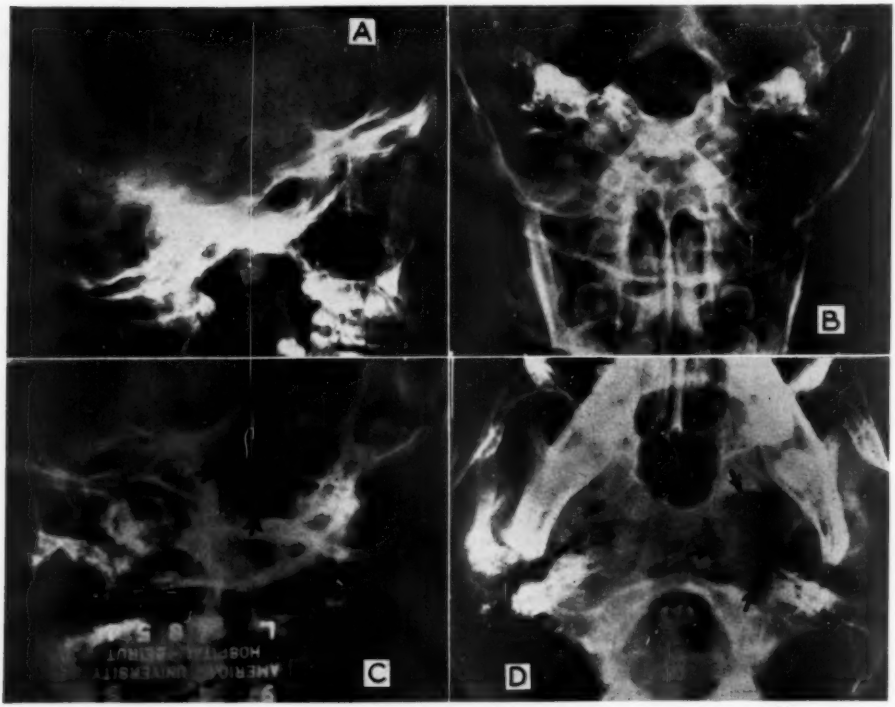

FIG. II. PRE-OPERATIVE RADIOGRAPHS OF SKULL.
(a) Lateral projection showing faint line of calcification (arrows).
(b) Towne's projection apparently normal.
(c) Stenver's projection showing destruction of the apex of the left petrous bone.
(d) Basal projection showing destruction of the left petrous apex.

Treatment and Progress

Her convalescence from this operation was straightforward. Deep X-ray therapy was started on the 7th post-operative day by Dr. Carter Braine, as follows: small-beam, directed radiation at 215 kV. and 15 mA. was used, with 1 mm. copper and aluminium filters. Left lateral, right lateral, and right posterior fields, each 4.0 cm. square, were employed. Twenty-one treatments were given over twenty-one days, the skin dose being 2,200r on each lateral field and 2,100r on the posterior field. The total tumour dose was 2,117r.

given through the same field as before. The skin dose was 925r to each field and the actual tumour dose 943r.

Eight months after operation she reported again. Marked improvement was now obvious. The abducens palsy had disappeared. Voluntary movements had returned on the left side of her face, and her left eye now watered as well as her right. Hearing in the left ear had returned almost to normal by audiometry and her left labyrinth responded to caloric stimulation. X-rays showed increased calcification of the tumour and a slight

diminution in its size (Fig. III). A clinical report and X-rays from Professor Idriss fourteen months after operation indicated that her improvement had been maintained. The shell of the tumour was densely calcified while the centre appeared translucent.

FIG. III. RADIOGRAPHS TAKEN EIGHT MONTHS AFTER OPERATION AND DEEP X-RAY THERAPY.
(a) Lateral projection, and (b) Towne's projection for comparison with Fig. II. The tumour has calcified and its upper extent become apparent.

DISCUSSION

This patient thus presented with a set of cranial nerve palsies of rapid onset, but not accompanied by fever or by cachexia. The involvement of Vth, VIth, VIIth and VIIIth nerves on one side could be readily correlated with the area of bone erosion shown in the X-rays. It is noteworthy that the X-ray changes were not demonstrated by views which are commonly regarded as routine, but only by the basal projection and by the Stenver's oblique projections for the petrous bone. Without these particular projections the diagnosis would have remained uncertain, and conditions as far apart as polyneuritis and pontine glioma would have required consideration.

Prior to operation several diagnoses were entertained on the basis of the X-ray findings. Sarcoma, the commonest tumour of the temporal bone, seemed unlikely because of the robust condition of the patient. The absence of inflammatory features ruled out osteomyelitis. Osteoclastoma was considered as a possibility, but the distinction from a cholesteatoma of the petrous bone, which is the commonest benign tumour of this region (Proctor and Lindsay, 1947), caused us the greatest difficulty. These latter tumours, which are really inclusion dermoids, in distinction from the masses of epithelial debris often found in chronic otitis media, commonly present with a slowly progressive and painless facial palsy (Jefferson and Smalley, 1938; Pennybacker, 1944). They usually also present with a severe degree of deafness, but the VIth cranial nerve is not involved, as both in Jefferson and Smalley's and in Pennybacker's cases the cholesteatoma cavities remained confined to the petrous bone. Osteoclastomata on the other hand can spread to involve neighbouring bones and this may be an important point of distinction. Otherwise the X-ray appearances of bone destruction may be indistinguishable.

Osteoclastoma is usually regarded as a benign tumour with a good prognosis after treatment. Malignant forms, however, sometimes occur (Russell, 1949). Complete removal can usually be accomplished only in such situations as a rib, the fibula, or mandible. Elsewhere in the body it has been found that incomplete removal is likely to be followed by recurrence, and the consensus of opinion is that it is preferable not to curette these tumours when complete removal is impossible, but to give deep X-ray therapy (Prossor, 1949; Ellis, 1949). Comparatively modest doses of radiation are recommended, for example 2,000r-3,000r

over twenty to thirty days. In favourable cases there is a temporary increase in the size of the tumour and further decalcification. After about two months recalcification commences and the tumour gradually becomes smaller and surrounded by dense bone. Often the normal architecture of the bone is not reformed and cystic spaces remain at the tumour site. With radiation therapy along these lines there is an 80-90 per cent. prospect of permanent recovery (Prossor, 1949; Ellis, 1949).

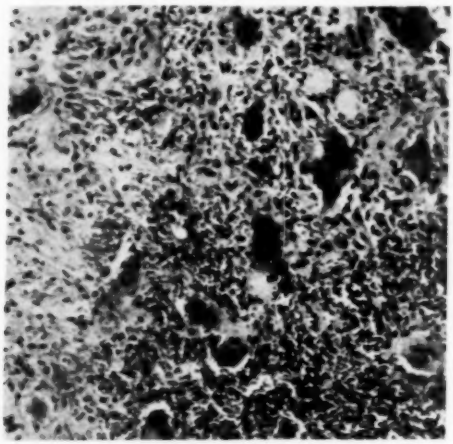

FIG. IV. Photomicrograph of tumour. (x 120)

In the literature 3 cases have been reported in which the petrous bone was the site of an osteoclastoma (Doderlein, 1913; Ramadier and Tournay, 1937; Lord and Stewart, 1943) and in a further case, both the mastoid and petrous bones were involved by an osteoclastoma (de Winter, 1952). Thus 5 cases are available for comparison (Table 1). All were females and their ages ranged from 8 to 50 years. The principal initial symptoms were pain (3 cases) and either deafness, vertigo or tinnitus (3 cases). Deafness appeared at some stage in every case while vertigo was noted in the 4 earlier cases but not in ours. Other cranial nerves commonly affected are the VIIIth (3 cases), the Vth (3 cases), and the VIth (2 cases). Apart from the earliest case (in which presumably no X-rays were taken) well marked X-ray changes of bone erosion were observed in each instance, thereby focussing attention on the temporal bone. In each case the diagnosis was established by biopsy. Subsequently four cases were treated by radiation therapy, two with apparent cure, and two with remission of symptoms of 10 and 14 months respectively before dying of extensive local recurrence.

The variations in age incidence and response to treatment of these osteoclastomata of the petrous bone are paralleled by the range of variation found in larger series of osteoclastomas arising in more typical sites. Presumably, therefore, the therapeutic problems of osteoclastomata in general are applicable to these same tumours occurring in the temporal bone. Removal of the tumour was attempted in 3 cases but was not successful, presumably because of technical difficulties. It is of interest to note that the two patients who ultimately succumbed in spite of radiation therapy had both had extensive partial removals before being treated by deep X-rays. Surgical intervention should therefore be limited to obtaining biopsy specimens only. The results of radiation therapy in this case, so far, have been gratifying. The child has remained well and the cranial nerve palsies are recovering. Follow-up X-rays show that the tumour is steadily recalcifying and becoming smaller. The prospects of complete recovery seem excellent.

SUMMARY

A case of osteoclastoma of the petrous temporal bone in an 8 year old girl is reported. She presented with involvement of the VIIIth, VIIth, VIth and Vth cranial nerves, of rapid onset but without constitutional disturbance. The clue to diagnosis lay in the X-rays of the temporal bone which showed virtual "amputation" of the petrous apex. The nature of the tumour was ascertained by biopsy, and treatment by deep X-ray therapy has resulted in arrest of the lesion.

Comparison of this case with four previous cases reported in the literature, shows that the commonest symptoms of this condition appear to be deafness, vertigo, pain, and facial palsy. These tumours clinically have to be distinguished from cholesteatomata (epidermoids) of the petrous bone, and are probably best treated by biopsy followed by deep X-ray therapy.

TABLE 1

COMPARISON OF FIVE CASES OF OSTEOCLASTOMA OF THE PETROUS BONE

Case	Clinical Features										Treatment	Result
	Sex	Age	Onset	Initial symptom	Deafness	Vertigo	Tinnitus	Pain	Other cranial nerve lesions	Pyrexia	X-ray signs		
1.—(Doderlein)	F	37	Gradual	Pain	Yes	Yes	Yes	Yes	No	No	—	Partial removal	Death one month after operation (temporal lobe abscess)
2.—(Ramadier and Tournay)	F	36	Gradual	Pain	Yes	Yes	Yes (violent)	Yes	VII, V (doubtful)	No	Yes	Biopsy followed by radiotherapy	Cured
3.—(Lord and Stewart)	F	50	Gradual	Tinnitus	Yes	Yes	Yes	No	No	No	Yes	Partial removal followed by radiotherapy	Death two years after operation from extensive local spread, after remission of ten months
4.—(de Winter)	F	35	Gradual	Pain	Yes	Yes	No	Yes	VII VI, V	No	Yes	Removal (twice) and radiotherapy	Remission of fourteen months following deep X-ray therapy. Malignant change two years after onset. Death three years after onset from extensive local recurrence
5.—(Present Case)	F	8	Rapid	Diplopia	Yes	No	No	No	VII, VI, V	No	Yes	Biopsy followed by radiotherapy	Apparent cure after one year

ACKNOWLEDGEMENTS

I wish to thank Mr. Murray A. Falconer, M.Ch., F.R.C.S., Director, Guy's-Maudsley Neurosurgical Unit, for assistance and advice in the preparation of this paper. Professor H. Idriss kindly supplied the pre-operative Stenver's projection. The photographs were taken by Mr. C. E. Engel, A.R.P.S., A.I.B.P., Department of Medical Illustration (Photographic), Guy's Hospital.

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CHORDOMATA

A REVIEW OF NINETEEN CASES OF CHORDOMATA INCLUDING FIVE VERTEBRAL CASES

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A CHORDOMA is a neoplasm of peculiarly specialized type arising in the region corresponding to the course originally taken by the foetal notochord, being found most commonly at the caudal and cephalic ends of the human spinal column. It is still considered to be somewhat rare, less than 300 cases having been described to the end of 1950; of these, 8 have been recorded in Australia (Newland and Woollard, 1929; Willis, 1930, 1933, 1948; Cato, 1933). A further case of special interest has recently been recorded by Maynard (1953).

In this paper are collected 19 examples of chordoma, the material being drawn from Melbourne, Sydney, Adelaide and Launceston over the last 25 years. These will be presented as a clinico-pathological study preceded by a short discussion of the embryology of the notochord and presumed histogenesis of the chordoma.

HISTORICAL

In 1856 Luschka described small masses of soft jelly-like tissue arising by a fine pedicle from the surface between the *sella turcica* and *foramen magnum* inside the cranium. Virchow (1857) attributed these masses of tissue to degeneration of cartilage calling them "*ecchondrosis physaliphora*"; the second portion of this name is descriptive of the vacuolated, "bladder"- or "bubble"-like character of the cells.

Müller in 1858 ascribed their origin to notochordal tissue but it was not until 1894 that Ribbert adduced experimental evidence to support Müller's hypothesis. He punctured the inter-vertebral discs of rabbits with resulting herniation of the *nucleus pulposus*. The tissue which was extruded grew and, histologically it closely resembled the tumour which was given, by Ribbert (1894), the name chordoma. This experiment has been repeated recently with a similar result by Congdon (1952).

Although until recently most attention has been directed towards the sacral tumours a cranial chordoma was first described in 1907 by Fischer and Steiner, whereas the first sacro-coccygeal chordoma was described in 1910 (Feldmann). Chordomata of the vertebral column have been well recognized only since the publication of Syme and Cappell's case in 1926, although one at least had been described before then (Fabricius-Möller, 1919).

It is not proposed, however, to review the literature in detail since there are many good reviews available, notably those of Stewart (1922), Stewart and Morin (1926), Cappell (1928) and Mabrey (1935).

Embryology of the notochord

The notochord originally came into existence when the axial symmetry of the vertebrates replaced the radial symmetry of the invertebrates. In the embryo it develops as a solid cord of cellular and, later, mucoid tissue, lying ventral to the neural canal and extending the whole length of the body. As the embryo develops, mesodermal tissue surrounds the notochord and from this surrounding tissue the vertebrae and intravertebral discs arise — the notochord thus being sited within the bodies of the vertebrae and the inter-vertebral discs. The vertebral portion then disappears in the main, notochordal tissue being represented in the *nucleus pulposus* of the intervertebral disc.

Early in its development the notochord is a hollow tube but it then becomes a solid cord of indifferent polygonal cells surrounded by a hyaline sheath. Later these cells become vacuolated.

These vacuoles contain a gelatinous mucin-like substance which later escapes from the cells forming a mucinous matrix in which lie syncytium-like groups of cells. Embryological features of the early stages of

development show that the notochord is then in close relation to the gut so that its making contact with or even entering the tissues of the pharyngeal vault is understandable. This relation has been emphasized by Cappell (1928).

HISTOGENESIS OF CHORDOMA

It is stated (Horwitz, 1941) or tacitly assumed (Willis, 1948) that chordomata arise from notochordal rests. Although there appears no doubt that tissue resembling notochordal tissue may be found in the adult human being in sites other than the inter-vertebral discs, to say that chordomata arise from this notochordal tissue is to return to Cohnheim's cell-rest hypothesis which has been rejected by most pathologists of our day. One might expect proliferating tissue in the vertebral region to differentiate in the direction of a mucoid tissue resembling the notochord. Indeed, such a differentiation is seen in the cells of the *annulus fibrosus* in cases of prolapsed disc. In such cases, cells lying in the *annulus* and in the "prolapsed" portion and which can be shown to be due to metaplasia of connective tissue cells, are often morphologically indistinguishable from typical physaliphorous cells. Therefore, we do not need to postulate an origin from specific cells in the case of these tumours. Cohnheim's cell rest theory to-day is an hypothesis for which, despite innumerable studies, we have no evidence and indeed considerable evidence leading to a different view has been accumulated.

With increasing recognition of the multipotent properties of cells (Nicholson, 1922; King, 1952) it has become manifestly impossible to state definitely the origin of a given mass of neoplastic cells. Hence, when we apply the term chordoma to a tumour we mean that structurally it resembles notochordal tissue and though it is conceivable that it may have arisen from true notochordal remnants, it is as, or even more, likely (as far as present evidence goes) to have developed by a process of differentiation from any of the tissues which replace the notochord. This is a special example of the use of the histological as oppossed to the histogenetic naming and classification of tumours.

Classification

In the original definition it was stated that a chordoma may arise at any point along the spinal column and this means that it may occur at any point from the pharynx to the coccyx. It is a matter of practical experience that this actually occurs but the common sites of origin are those of the cranial and sacro-coccygeal areas.

Coenen (1925) produced a classification which is still of practical use.

(1) Cranial	(a) sphenoid.
	(b) clivoid.
	(c) naso-pharyngeal.
(2) Vertebral	
(3) Sacro-coccygeal	(a) ante-sacral.
	(b) sacral.
	(c) retro-sacral.

The occurrence of tumours not only in, but adjacent to the sphenoid bone is an important observation as far as the conclusions regarding histogenesis (stated above), are concerned. It may be that, for example, the naso-pharyngeal tumour is merely an extension of a sphenoidal growth but the very fact of differentiating the former exemplifies the point of view adopted here that all we are justified in accepting in the present state of our knowledge is the presenting character of the growth. We have insufficient evidence on which to build an edifice of precise mode or site of origin.

TABLE 1

Site incidence of chordomata as recorded in the literature to the end of 1950 (present series in parentheses)

Cranial	105	(1)
Vertebral	36	(5)
Sacro-coccygeal	143	(13)

A total of 284 chordomata has been recorded to the end of 1950 the site distribution being shown in Table 1. The age distribution is widespread and no age can be said to be exempt. The mean age in the present series excluding the single cranial case was 54. Generally speaking the greatest

incidence of sacro-coccygeal cases is in the 5th and 6th decades whilst the cranial group has its maximum incidence in the 4th and 5th decades. This difference in age incidence is probably explained on the basis that the cranial cases show earlier symptoms and kill earlier by virtue of their site rather than the age incidence being different *per se*. The sex incidence shows a slight preponderence of males in this series and most others, raising the question of the part played by trauma in the aetiology of these tumours. In most series the proportion of cases with a history of trauma is small and often the trauma antedates the tumour's appearance by many years. Details of the present series are recorded in Table 2.

FIG. I. Photograph of patient (Case 17) with a large chordoma of the sacrum showing the condition in October, 1952. Some increased pigmentation may occur in the intact skin over the tumour. Despite the large size there are no symptoms of incontinence.

MORPHOLOGY OF THE TUMOUR

Macroscopically the tumour when seen at operation or autopsy is usually large in size (it may be many inches in diameter), in general is spherical in shape, is variegated in colour and firm in consistency. It is apparently encapsulated although local invasion of adjacent structures may be obvious. One of the important features is the destruction of bone which it replaces.

FIG. II. Skiagram of the sacrum of a patient with a chordoma (Case 7) illustrating destructive and expansive growth. There is a large, soft tissue shadow extending laterally on the left of the coccyx (which is viewed from the front).

On cut section the tumour is again variegated in appearance. It is lobulated, gelatinous in consistency, greyish to pink in colour and frequently shows haemorrhagic areas (Fig. III). Necrotic areas may be seen and occasionally cysts filled with jelly-like material. Often the tumour has a quite well-defined capsule. The important feature of bone destruction is best appreciated by examination of X-ray films (Fig. II).

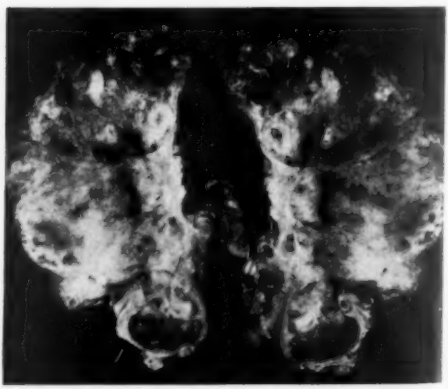

FIG. III. Photograph of a macroscopic operation specimen of sacro-coccygeal chordoma (Case 8). The gelatinous type of tissue, apparent encapsulation and areas of haemorrhage are all well illustrated.

TABLE 2

Tabulated summary of clinical details of 19 cases of chordoma

No.	Sex	Age	Presenting Symptom	Duration	Site	Trauma	Treatment	Operative and/or necropsy findings	End result
1	M	73	Lump R side sacrum; pain R leg recently	5 years	sacrum	no	Partial excision: deep therapy	Operation:—soft yellowish lump eroding bone at sacro-coccygeal junction	Alive 6½ years after first symptom
2	F	66	Pain in rectum with constipation; recent loss of weight	4 years	sacrum	no	nil	Autopsy:—sacrum replaced by vascular globular projecting mass 4" in diameter	Death 4 1/12 years after first symptom
3	M	57	Urinary frequency; recent pains in legs	5 years	1st lumbar vertebra	Sciatica 30 years previously	Laminectomy: deep therapy	Operation:—dura bulged irregularly over 12T and 1L vertebrae. Cord surrounded by dark tissue	Death 8½ years after 1st symptom
4	F	49	Nagging pain, lower back; recent lump over sacrum	2 years	sacrum	no	Excision: deep therapy	Operation:—soft encapsulated tumour eroding sacrum and invading both buttocks. Autopsy: no recurrence	Death 4½ yrs. after first symptom (aplastic anaemia)
5	F	45	Nagging pain, lower back	14/12	sacrum	no	Excision: deep therapy	Operation: soft vascular tumour 3" diameter invading sacrum	Death 6 yrs. after first symptom
6	M	55	Nagging pain, rectum radiating to buttocks; recent constipation	6/12	sacrum	no	Excision: deep therapy	Operation: gelatinous friable tissue eroding anterior aspect sacrum	Alive 6½ yrs. after first symptom
7	M	67	Pain, lower back	6/12	sacrum	yes 6/12 before	Deep therapy after biopsy	Aspiration biopsy only	Alive 2 yrs. after first symptom
8	F	54	Pain at base of spine	2 years	sacrum	no	Excision	Operation: vascular tumour brain-like in consistency eroding sacrum	Death 6 yrs. after first symptom
9	F	63	Pain across shoulders	2 years	4th and 5th cervical vertebrae	no	nil	Autopsy: 4th and 5th C vertebrae replaced by soft white friable tumour tissue which bulged pharynx	Death 2 1/12 yrs. after 1st symptom
10	F	63	Diplopia	2 years	cranial	no	Partial excision	Operation: firm white fleshy tumour tissue to left of pituitary area: it spread up and back	Death 8 yrs. after first symptom
11	M	51	Pain R shoulder and back; recent paraplegia	18/12	7th cervical 1st and 2nd thoracic vertebrae	no	Laminectomy: deep therapy	Operation: firm gelatinous tumour invading spines and arch of C7 T1 and T2 with body of T1 affected	Death 4 yrs. after 1st symptom with massive recurrence in muscles of back
12	M	62	Lump, L buttock then L groin	12/12	sacrum	no	Excision	Operation: necrotic haemorrhagic tumour 3½" diameter excised. Recurrence in 6/12 and reoperated	Not traced
13	M	34	Pain back and weakness, R leg	6/12	2nd lumbar vertebra	no	Laminectomy: deep therapy	Operation: biopsy taken and diagnosed histologically secondary from carcinoma of kidney. Readmitted later with lump R loin when correct diagnosis made	Death 5½ yrs. after first symptom
14	M	63	Lump, R buttock	2/12	sacrum	Heavy fall 6/12 previous	Excision	Operation: large mucoid tumour 3" diameter eroding sacrum and ilium	Not traced
15	F	33	Pain, lower back	12/12	sacrum	no	Excision	Operation: small tumour mass adherent to upper portion of coccyx	Alive and well 9 years after first symptom
16	F	35	Pain, lower back; local swelling present 2/12	3½ years	sacrum	no	Deep therapy after biopsy	X-ray: destruction of sacrum. Mass felt on rectal examination	Death 12 yrs. after first symptom
17	M	65	Lump over sacral area	10 years	sacrum	no	Deep therapy after biopsy	Autopsy: large tumour size 7"x7" weighed 7 lb. mainly recto-sacral: death due to myocardial infarction and carcinoma of the lung	Death 14 yrs. after first symptom
18	M	61	Pain, lower abdomen and hips	4½ years	sacrum	no	Partial excision deep therapy	Operation: large mass growing round rectum from sacrum	Alive 5½ yrs. after first symptom
19	F	44	Pain, back of neck; recent quadriplegia	3/12	1st and 2nd cervical vertebrae	no	Laminectomy	Operation: no tumour seen. Autopsy: tumour found outside dura below foramen magnum compressing cord. Like stiff mucus in consistency	Death 9/12 after first symptom

Histologically, the tumour is characterized by a clear demarcation between parenchyma and stroma but there is a very wide variation in the appearance of the parenchymal cells. This variation is due, for the most part, to the presence of mucin in greater or lesser degree so that we can grade the tumours histologically into 4 types:

(1) Cellular
  (a) carcinomatous type.
  (b) sarcomatous type.
(2) Cellular with physaliforous cells.
(3) Cellular with extra-cellular mucin.
  (a) cords of cells.
  (b) small masses of cells.
  (c) "early notochordal" structures.
(4) Grossly mucoid.

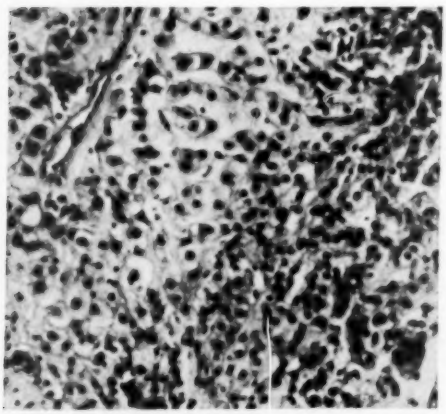

FIG. IV. Photomicrograph of a section of the tumour from Case 1. It shows a very cellular portion on the right while mucin production is in evidence round the vessel in the figure. (x 150)

These groupings indicate only what is a predominant appearance since in the one tumour it is possible to find gradations from a cellular hyperplastic tissue to tissues burdened with mucin. The importance of the classification arises in the atypical examples when a diagnosis from a carcinoma or sarcoma on the one hand and a mucoid carcinoma on the other may be difficult.

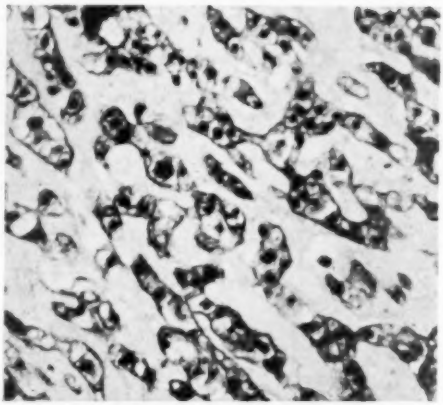

FIG. V. Photomicrograph of a section of the tumour from Case 1, showing extensive mucin production with many cells distended with mucin much of which is extra-cellular. (x 150)

The carcinomatous type is seen in Fig. IV where the cells are of epithelial type, the cell outlines being indistinct in one part so that the tissue is almost of a syncytial character. Further to the left in this figure the cells may be seen to be polygonal in shape with clearly defined margins while mucoid material is present in both intra- and extra-cellular vacuoles. There is considerable variation in cell size. Where mucin is present in large amounts the cells appear to become shrunken whilst the nuclei are darker in staining reactions (Fig. V).

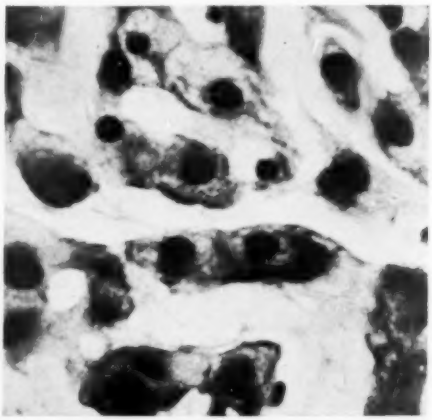

FIG. VI. Photomicrograph of a section of chordoma (Case 1) showing cords of epithelium-like cells, the cords being separated by mucin which appears as precipitated strands. (x 1050)

The mucin varies directly in amount with the length of the life history of the cell. At first the mucin droplets are minute and intra-cellular but with continued growth the vacuoles enlarge considerably and some mucinous collection appears outside the cell membrane forming a characteristic inter-cellular accumulation. This breaks the cellular aggregations up into cords (Fig. VI) and into groups of shrunken cells isolated by relatively large amounts of mucin (Fig. VII).

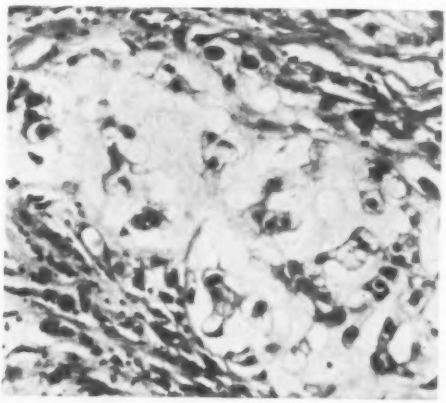

FIG. VII. Photomicrograph of section of a chordoma (Case 8) showing a lobule of physaliphorous cells with extensive mucin production. The lobule is surrounded by a hyaline capsule which is infiltrated by small round cells. (x 150)

The diagnostic feature of these tumour cells is the presence of both intra- and extra-cellular mucin producing the "bubble"- or "bladder"-like appearance the feature which originally gave rise to the term (Fig. VIII). Nuclear vacuolation is seen in this figure. Large multinucleated cells are common and these are always distended with mucin (Fig. IX). The mucin occasionally is concentrated on one side of the cell compressing the nucleus or producing a signet ring appearance (Fig. X), similar to those found in carcinomata of the stomach and bowel.

The stroma generally is fibrous but in certain areas is hyaline—this occurs particularly in the vertebral chordomata. Quite commonly focal accumulations of small round cells are to be found in the stroma (Fig. VII).

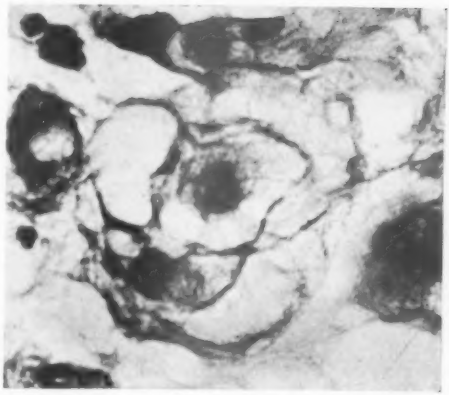

FIG. VIII. High power photomicrograph of section of a chordoma showing typical physaliphorous cells. One nucleus shows a vacuole. Most vacuoles are present within the cells but others are seen as bladder-like extensions outside the cells. (x 1050)

The tumour is very vascular, both old and recent haemorrhages being common. The main vessels run in the stroma but much blood is contained in sinusoidal-like cavities among the parenchymal cells.

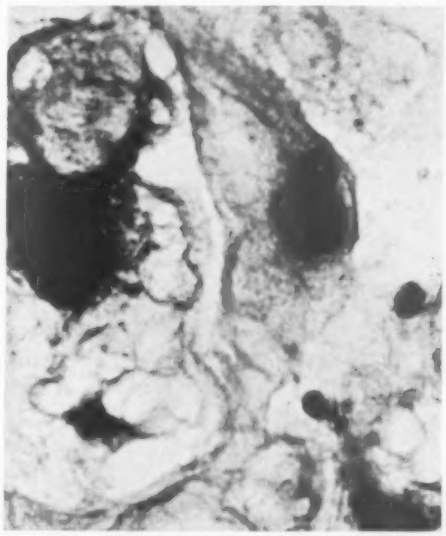

FIG. IX. High power photomicrograph of a section of a chordoma showing multinucleated giant cells distended by a very large amount of mucin. (x 1050)

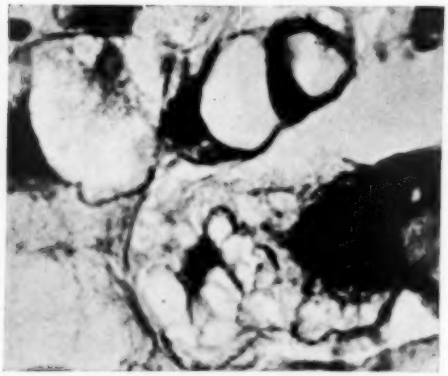

FIG. X. High power photomicrograph of a section of a chordoma showing several cells with signet ring form adjacent to typical physaliphorous cells. (x 1050)

X-RAY APPEARANCES

The radiological signs of sacro-coccygeal chordomata were stated by Hsieh and Hsieh (1936) as:

(1) Expansion of the tumour mass. This may be demonstrated in both antero-posterior and lateral views.

(2) Rarefaction or destruction. The involved bone presents either a loculated appearance, with multiple small circular or oval radio-translucent areas or the bone is destroyed over a large area.

(3) Trabeculation. The remains of the undestroyed bone form dense trabeculae which may extend into the soft tissue mass outside the original normal boundary of the bone.

(4) Calcification. This may be due to reactive new bone formation or deposits of calcified material in the tumour following degeneration or necrosis.

Most of these radiological features can be seen in Fig. II.

Hsieh and Hsieh add that these appearances may be found to some degree when the tumour is situated in the vertebral column or cranial cavity.

SYMPTOMATOLOGY

There is no symptom characteristic of chordomata as a group but pain, or, in the case of cranial tumours, headache is a constant concomitant at some stage of the disease. In the cranial cases headache and visual disturbances are the most common symptoms while the signs are those comparable to most intra-cranial neoplasms. In the vertebral cases pain followed sooner or later by paralysis of lower motor neurone type, the limbs affected depending on the level of the vertebra involved, is the most common symptom sequence. In the sacro-coccygeal cases pain is a universal symptom and it was present in all cases in this series.

Case 17 (Fig. I) is somewhat unusual in that the patient complained only of a dull ache if he sat for any length of time. The pain in these cases varied from a dull nagging ache to "sciatica," its nature depending on the anatomical relation of the tumour to the nerves in the vicinity. The early association of pain with a tumour is found particularly in the retro-sacral type. The tumour may be extraordinarily large and yet occasion relatively few symptoms as in Stewart and Morin's (1926) case and in Case 17 of the present series. Constipation is common, together with urinary disturbances, in the later stages. Stewart (1922) remarked on the lack of ulceration of the skin produced by the tumour while Mabrey (1935) stated that the tumour often ulcerated the rectal mucosa but such an event did not occur in any case in this series.

DIAGNOSIS

In practically all cases the diagnosis depends ultimately on an histological examination of a section of the tumour, the diagnosis being clinched by the finding of typical physaliphorous cells.

Clinically the cranial group cannot be differentiated with certainty from any other intra-cranial neoplasm, whilst in vertebral cases metastases from a distant primary source must be considered as well as diseases such as multiple myelomata. Prolapsed inter-vertebral disc should also be considered and it is interesting to note that one case of "chordoma," diagnosed histologically which was in reality a prolapsed

disc, was found during the collection of the present series. The case occurred in 1932 when the last mentioned condition was only then becoming recognized. At operation a small cherry-sized tumour was found between the 4th and 5th lumbar vertebrae, being connected to the inter-vertebral disc by a pedicle. The patient so far as is known is alive and well to-day while the histological section is that of a herniated disc. Case 13 of this series was diagnosed first as a metastasis from a Grawitz tumour, a diagnosis which was later corrected.

Sacro-coccygeal cases are perhaps more easily diagnosed clinically but even here, uterine fibroids, carcinoma of the rectum, dermoids and bone tumours including primary sarcoma as well as metastases from other sites in the body must all be considered in the differential diagnosis. The writer recently performed a necropsy where a very small carcinomatous ulcer of the bladder, 1 cm. in diameter had grown posteriorly round both sides of the rectum and had then completely replaced the sacrum. Apart from a few small metastases in the liver there was no other demonstrable tumour focus and such a case may present a diagnostic problem.

TREATMENT

The condition, in so far as cranial and vertebral cases are concerned, carries a hopeless prognosis but where surgical removal is at all feasible operation offers the only hope of cure. This may have occurred in Case 15 where the growth was localized to the upper end of the coccyx and also in Case 4 where death was due to aplastic anaemia following radio-therapy. At necropsy no recurrence was demonstrable in this last case. However, because of the size and extent of the growth and the involvement of adjacent structures, complete removal and therefore cure is not possible in the majority of sacro-coccygeal cases. Relief of untoward pressure symptoms by partial removal is certainly indicated where possible, even if multiple operations be required (Faust *et alii*, 1944).

The value of deep therapy, both with and without operation is difficult to assess. There is no doubt that it does alleviate pain—one patient (Case 16), for example, would disappear for six months at a time and then reappear to ask for more therapy—so that it is worthwhile even in advanced cases.

DISCUSSION

On examination of the extensive literature of chordomata one finds that the relative proportion of cranial to vertebral chordoma is as 3:1 but, in the present series this ratio is more than reversed indicating that vertebral chordomata are perhaps not so rare as has been thought in the past. In fact, where there is a localized vertebral lesion without an obvious primary site chordoma should be considered in the differential diagnosis.

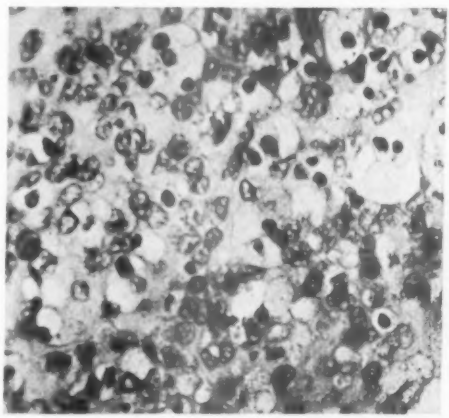

FIG. XI. Photomicrograph showing portion of a section from a very cellular chordoma (Case 18). Mucin production is small in amount although a few typical physaliphorous cells may be seen. (x 350)

A chordoma is a malignant tumour showing the fundamental properties of continued and invasive growth; it is, moreover, a malignant tumour which occurs in sites where, in the majority of cases, radical removal is not possible, at least in that stage when patients usually first present. However, it is, as will readily be seen from Table 2, generally slowly growing. It does, however, kill relatively quickly in those sites where expansive growth is limited early by vital structures such as in the cranium and vertebral column.

There is no doubt that both sacral and vertebral cases metastasize and reports of this occurrence are relatively common in the

literature. Local invasion, however, was the predominant feature of practically all the cases in the present series and no definite case of metastasis was found. In Case 11, a secondary mass made its appearance on the right shoulder probably as a result of lymphatic spread; this was in addition to massive recurrence at the primary site in the 1st thoracic vertebra. In this series 12 deaths have occurred so far but in only 5 cases has necropsy been performed so that true visceral metastasis cannot be excluded. Mabrey (1935) notes that in recorded necropsies on cases of sacro-coccygeal chordoma metastases were present in 62.5 per cent. of cases.

Maynard (1953) has recently described a case, occurring in Melbourne, of a lumbar chordoma with a pulmonary metastasis: it is noteworthy that the necropsy in this case was performed by Professor M. J. Stewart who first drew the attention of English pathologists to this tumour. There is as yet, so far as one can ascertain, no record in the literature of a cranial chordoma producing distant blood-borne metastases. The cranium is also the only site where the benign *ecchondrosis physaliphora spheno-occipitalis* of Virchow is to be found. This is a symptomless, small, flattened, jelly-like nodule arising by a narrow pedicle from some part of the clivus. It is claimed that they occur in 2 per cent. of necropsies. They are hamartomata or malformations and may be designated, if there be any virtue in the term, as "notochordal rests" similar to those described microscopically in the sacrum and vertebrae by Horwitz (1941) and other authors. Whether these "rests" are present and do give rise to the true chordoma is the debatable question discussed above.

The macroscopic appearance of the chordoma appears to be similar in all sites namely, a gelatinous type of tissue apparently encapsulated, with numerous haemorrhagic areas and showing a propensity for local invasion and destruction comparable, as Stewart (1922) points out, with the behaviour of the basal cell carcinoma of the skin. Microscopically the appearance varies considerably from extreme cellularity with minimal mucin production (so that it is difficult to find true physaliphorous cells) in which case the tumour is liable to be labelled sarcoma or secondary carcinoma (Fig. XI), to a relatively acellular type with an extraordinarily large amount of mucin. This last appearance is, in the present series, a predominant feature of the vertebral types and this observation does not appear to have received emphasis previously. The vertebral types produce approximately circular aggregates of cells with extreme mucin production, surrounded by a thin layer of hyaline tissue (Fig. XII), but nothing comparable with Cappell's "notochord structure" was seen. The sacro-coccygeal types, however, commonly show a number of appearances in the one histological section.

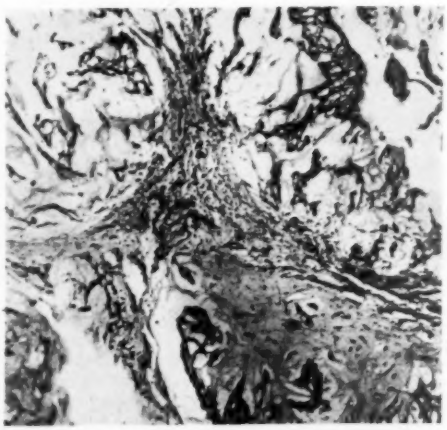

FIG. XII. Low power photomicrograph to show the typical histological appearance of a vertebral chordoma (Case 19). Extreme mucin production is seen in lobules which have a hyaline connective tissue capsule.

Nuclear vacuolation was occasionally seen in the more cellular tumours but was never a gross feature. Mitotic figures were rare but again were more common in the cellular tumours. The cellular morphology showed great variation not only with regard to mucin content but also in size, shape, number and staining characters of the nuclei. The mucin presented a wide range of appearances being granular in one portion of a section, reticulated in another and homogeneous in another. Often it had disappeared (perhaps from contraction during fixation which was by formalin in all cases), leaving empty spaces. Where the mucin formed a

homogeneous mass the resemblance to cartilage on superficial inspection was pronounced.

The clinical diagnosis in all cases is usually one of exclusion but the value of aspiration biopsy should be remembered. This applies particularly where, at first presentation, the growth is so advanced that no radical procedure is advisable, the diagnosis being clinched by the finding histologically of typical physaliphorous cells.

SUMMARY

Nineteen cases of chordoma are described, the series being composed of 1 cranial, 5 vertebral and 13 sacro-coccygeal cases. The vertebral type, therefore, was found, in this series, to be more common than was formerly believed.

The tumour histologically resembles notochordal tissue, both in its cytological appearance and in its production of mucin, but need not, in the light of present knowledge, be regarded as arising from notochordal rests. By analogy with more commonly observed tumours it can develop from any of the tissues which occur in the region of the notochord in the adult organism and which may produce mucin. Their structure and behaviour are discussed. The ultimate prognosis in the majority of cases, particularly the cranial and vertebral types, must be considered as poor. The radiological appearances, symptomatology and treatment are briefly considered. Pain is the almost invariable presenting symptom while palliative surgery and deep therapy both have their place in treatment.

ACKNOWLEDGEMENTS

The collection of this series of chordomata has only been made possible by the kindness of many of my senior colleagues who readily provided both clinical data and histological material.

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I am indebted to the clinical photographer of the Royal Melbourne Hospital, Mr. R. Ingles, for Figure I.

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HYDATID CYST OF THE KIDNEY

A REPORT OF TWO CASES

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Christchurch Hospital, New Zealand

THIS record is of two cases of hydatid infestation of the kidney, unfortunately not diagnosed until hydatid material was produced in the voided urine. Both patients were robust, active farmers in their mid-thirties and men who had spent their lives in the country. In each case considerable time elapsed and there was a fair amount of intermittent disability before a diagnosis was made. In both a cure by spontaneous evacuation of all the hydatid material from the kidney may have been achieved. The case histories of these two men will be described.

Case 1

In June, 1948, L.P. was admitted suffering from right renal colic. There was an insidious onset of dull, aching pain in the right flank at 8 a.m. He went to work and later the pain became a severe colic radiating from the loin to his testicle and he lay on the ground and vomited. There were no urinary symptoms and no frequency of micturition. He was able to make his way to a doctor but an injection of morphia proved unavailing and he was sent down to hospital. There had been three similar attacks within the last three years but they had all terminated within two hours. He had not passed anything unusual in the urine. His general health was otherwise good and he was a very fit, active man. On examination, there was no apparent enlargement of the right kidney but tenderness over the lower segment of the ureter. On the following day cystoscopy was performed and it was noted that the right ureteric orifice was congested and the efflux was scanty. A number 6 catheter obstructed at 8 cms. but a fine catheter passed and released a free flow of urine which contained scanty pus and red blood cells and was sterile on culture. The right pyelogram showed no abnormality.

A diagnosis of non-opaque ureteric calculus was made and as the patient was now quite well and anxious to return to his farm he was discharged. Two years later he was again admitted suffering from right renal colic. This had been intermittent for the past nine days and five days before admission he passed some blood in the urine accompanied by some "tissue." The right kidney was slightly enlarged and tender and again he was very sensitive over the lower ureteric segment. Three days after admission cystoscopy revealed an acute general cystitis—a poor, cloudy efflux on the right side—and again there was difficulty in passing the right catheter at about 8 cms. There was retention of cloudy urine in the right pelvis. The ureteric urine contained profuse pus cells and red cells. A search for tubercle bacilli and hydatid elements was negative. There was no growth on culture. Right retrograde pyelography showed a partial obstruction to the ureter just above the pelvic brim (Fig. IA). The pelvis and calyces and the ureter proximal to the obstruction were moderately dilated. The kidney was slightly enlarged but its contour was normal. An intravenous pyelogram carried out four days later showed no function on the right side indicating that the obstruction was still present. The total white cell count was 19,000 and included 26 per cent. of eosinophils.

A suspicion of hydatid infestation was aroused but both the Casoni and complement fixation tests were negative. The patient was again discharged this time to a follow up clinic. In twenty days he brought to the clinic a bottle containing typical laminated membrane which he had passed without pain. The report from the pathologist was "A thin walled cyst resembling a hydatid ectocyst measuring 2.5 cms. in diameter. Two smaller cysts and a ragged mass with an attached firm cellular portion were also received. The cellular mass may be a bunch of microscopic daughter cysts. Sections of all the tissue show many of the fragments to possess a definite laminated structure typical of a hydatid ectocyst."

The patient, now quite well, reluctantly consented to admission for further investigation. A mid stream specimen of urine revealed nothing of note. Both Casoni and complement fixation tests were now positive and the eosinophilia persisted. Physical examination did not show any enlargement of the right kidney. Cystoscopy revealed a normal bladder—right ureteric orifice normal with a good, clear efflux and the catheter passed without difficulty. In the ureteric urine there were scanty leucocytes, moderate red blood cells, amorphous urates and no hydatid elements. There was no growth on culture. Bilateral retrograde pyelography was again carried out. This included lateral and oblique films of both kidneys but there was no evidence to indicate the presence of a hydatid cyst. The pyelographic findings appeared to be quite normal (Fig. IB).

There did not seem any reason to interfere surgically with this kidney if spontaneous cure was a possibility. He was admitted again in six months. He had been perfectly well and cystoscopy, pyelography, and examination of the urine returned negative findings. The hydatid sensitization tests were still positive.

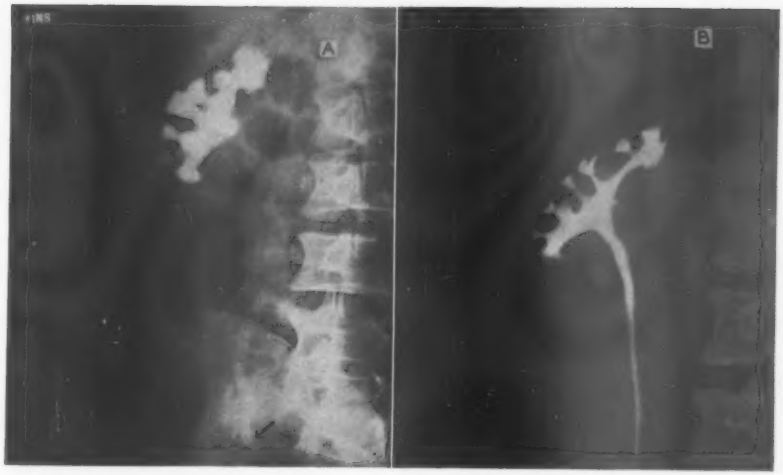

FIG. I. (A) Obstruction to the lower third of ureter with resulting pelvi-calycine hydronephrosis and hydro-ureter. (B) Three months later, normal pyelographic findings.

Case 2

The second patient was an active man of 35 years who had spent his life in the country amongst sheep and dogs. He was first seen in January, 1950, and gave a history of isolated attacks of left renal colic, associated with the passage of sand-like debris in the urine. These attacks had occurred over the past three years and he had sought advice but no investigation had been carried out.

When seen here he had just recovered from such an attack. Examination of the urine at this time revealed only amorphous debris and few red cells, and there was no growth of organisms. A plain film did not show any abnormality. Cystoscopy discovered a normal bladder mucosa, normal appearing ureteric orifices with a clear efflux and good function with dye on each side. A bilateral retrograde pyelogram was carried out. The right side was normal. The left renal shadow was not enlarged and appeared regular in outline. There was incomplete filling of the upper major calyx from an apparent constriction at the infundibulum of this calyx (Fig. IIA). Only a small amount of opaque material entered the calyx distal to the obstruction. The lower segment of the ureter was slightly dilated and there appeared in it some small filling defects. A ten minute emptying film showed some retention of dye in the affected calyx. Examination of the left ureteric specimen revealed red cells, scanty leucocytes and a search for tubercle bacilli was fruitless. There was no family or personal history of tuberculosis and in the intervals between attacks the patient was perfectly well without any urinary symptoms. A diagnosis was made of stricture of the infundibulum of the upper major calyx, and no further treatment was suggested.

He had another attack in three months time, passing some more sandy debris in the urine. The bout cleared up rapidly. In April, 1951, he again presented himself for examination after another attack and stated he had passed several minute stones in the urine. However, by the time he arrived in the city, the urine was again normal and he had not preserved any of the material voided. Cystoscopy did not discover anything abnormal but the left pyelogram showed very poor filling of the upper major calyx. The fill was repeated under pressure and for the first time a clear picture was obtained (Fig. IIB). There was some narrowing of the infundibulum of the upper major calyx. The upper minor calyx was normal but the lower minor calyx was ballooned and irregular, and evidently the seat of infection. It was not quite clear why, in a poorly draining upper unit, one calyx should be normal and the other distended and irregular.

The question of hydatid disease was raised at this stage but neither of the writers had ever seen a hydatid cyst attack a calyx except at its periphery and here the primary seat of obstruction was at the isthmus of the major calyx at its junction with the pelvis.

He was again discharged, this time with a bottle of saline solution and asked to preserve carefully any abnormal elements voided in the urine. In the interval there were several minor attacks but in March, 1952, he had a severe left colic and voided some "skin-like tissue" which he placed in his saline container and came at once to town. The receptacle held obvious hydatid material and the report on this was as follows:—"Several large and small fragments of thin hydatid material. Microscopic section shows the membrane to have the laminated acellular appearance typical of a hydatid ectocyst. Microscopic examination also reveals a number of hooklets and degenerate scolices."

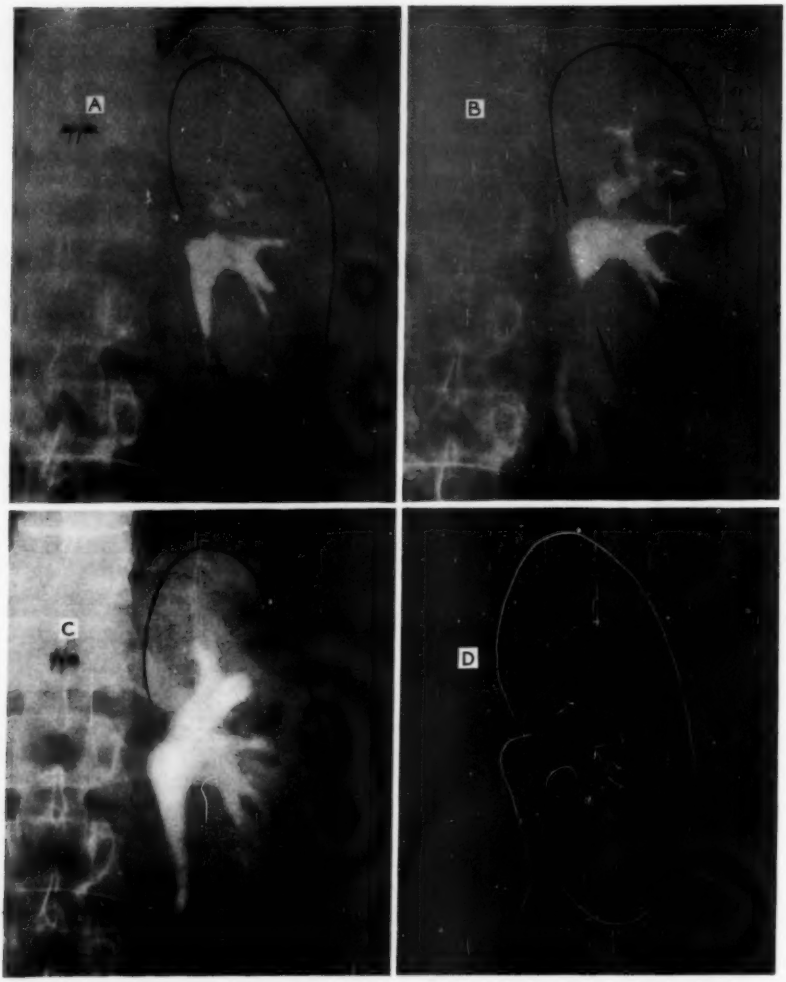

FIG. II. (A) Incomplete filling of the upper major calyx. (B) narrowing of infundibulum of upper major calyx with ballooning of its lower minor calyx. (C) Upper major calyx well filled but persistent ballooning of the lower minor calyx. (D) Line diagram showing para-pelvic position of the cyst and its site of rupture into the minor calyx.

At this time an intravenous pyelogram was carried out. The left upper major calyx was well filled. The upper minor calyx was normal but the lower minor calyx was ballooned yet regular in outline (Fig. IIC). There was a persistent irregularity on the supero-medial margin of the renal pelvis at its junction with the upper major calyx. No abnormality was found in the blood count—the Casoni test was positive and the complement fixation test was negative.

The evidence that was now available as a result of the serial pyelograms indicated that the cyst was situated in a para-pelvic position (Fig. IID). At the original examination in 1950 (Fig. IIA) the cyst was apparently filled with hydatid contents and was causing extrinsic pressure and obstruction to the infundibulum of the upper major calyx.

The appearances on the subsequent pyelogram in 1951 (Fig. IIB) indicated that partial evacuation of the contents of the cyst had occurred. The

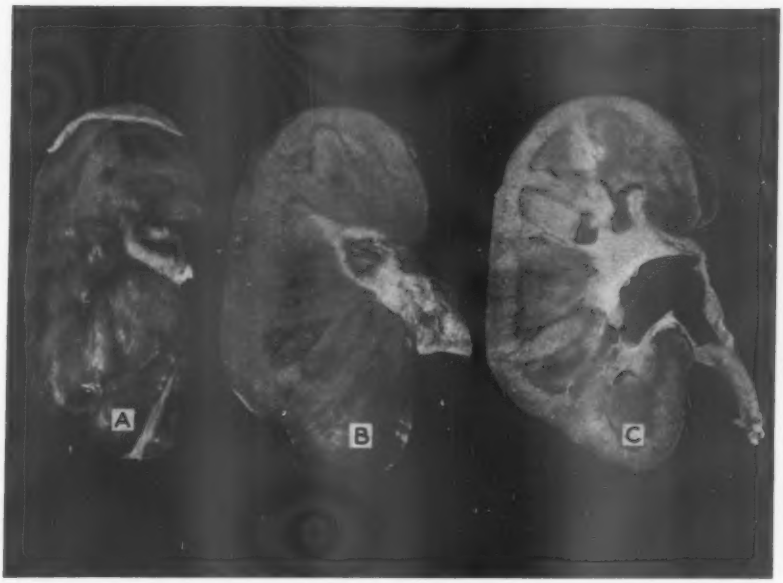

FIG. III. (A) Posterior surface of kidney showing cystic structure 2 cms. in diameter in the upper and outer part of hilum. (B) Section through the cyst showing its communication with a minor calyx. Note complete evacuation of cyst contents. (C) Deeper section showing the selective hydronephrosis of the postero-laterally directed minor calyx.

ballooning of the inferior minor calyx must have been produced by the selective rupture of the cyst into this calyx. Following removal of the kidney it was shown that the cyst was situated in a posterior para-pelvic position and that the affected minor calyx was directed postero-laterally (Fig. III). This would, therefore, account for its selective involvement. The final pyelogram (Fig. IIC), performed immediately after the patient had voided a considerable quantity of hydatid material, showed no evidence of obstruction to the superior major calyx. This was due to the evacuation of the contents of the cyst. The ballooning of the inferior minor calyx was still present and this was apparently due to calycine hydronephrosis resulting from the repeated discharge of hydatid material into this calyx.

The question then was, in view of this fairly complete emptying, whether anything further should be done. Experience has shown that complete emptying of all hydatid material from a renal cyst is rare and fractional emptying may continue over a period of many years. The patient, being in an isolated district, was anxious to be finally rid of this recurrent disability and removal of the left kidney was decided upon.

The operation was not difficult and few adhesions were found. Attention was directed to the upper pole where the cyst might be found but this appeared normal and the actual cyst was not discovered until the organ was removed.

PATHOLOGICAL REPORT

"Macroscopic.—The posterior surface of the kidney shows a firm, cystic structure 2 cms. in diameter lying in the outer and upper part of the hilum. On section, this cystic structure is seen to have a thick fibrous wall which is wrinkled internally and partly inverted. It communicates through a fairly narrow channel with an adjacent calyx (Fig. III). The cavity contains no obvious hydatid elements. The kidney substance appears otherwise normal.

Microscopic.—There are no viable hydatid elements in the section taken. This kidney shows the collapsed pericyst and some retained fragments of ectocyst of an *Echinococcus granulosus* cyst."

DISCUSSION

The sequence of events in the first case seems to follow the usual pattern of encroachment of a hydatid cyst on the urinary channels. First, the isolated attacks of colic due to the detachment of small, unrecognized pieces of the exposed laminated membrane of the cyst. Then a major attack of pain following rupture of the sac and the lodgment of the expelled contents in this case in the lower ureteric segment. The only pointer at this stage was the significant

eosinophilia. It was unusual that the smaller hydatid elements, hooklets and scolices could not then be identified in the ureteric specimen of urine, and that neither skin or serum test became positive shortly after the rupture. Judging from the size and extent of the expelled contents the cyst must have been the size of a large walnut, yet no trace could be found of its intrusion on the outline of pelvis or calyces. After rupture no sign was found of any sinus leading from the urinary tract.

In the second case there were, from the first, definite pyelographic findings which we were unable to interpret. The constriction of the infundibulum of the affected calyx seemed to favour more the "exclusion calyx" of tuberculosis. One generally expects a parenchymal lodgement of a hydatid cyst and encroachment on the calyx from the periphery. There was again exposure of the laminated membrane to the urinary stream, with the breaking off of small pieces and subsequent colic. From the description of the material voided the membrane was either lightly calcified or crusted with phosphates.

Then came rupture and apparently complete evacuation of all viable hydatid elements. A thorough search of the removed cyst failed to find any real residuum but the pericyst, and a small fragment of necrotic laminated membrane. A natural surgical cure had occurred which, in our experience, is unusual in the urinary tract. Operation was unnecessary in this instance. The other unusual feature was the site of lodgement of the cyst in the para-pelvic tissues. To reach this site the embryo had apparently made the circuitous journey through two complicated capillary beds firstly the pulmonary and secondly the renal circulations. It probably came to rest in the small pelvic tributary of the renal vein. The established cyst with plenty of space in which to expand in the para-pelvic tissues then chose to encroach on the renal substance and actually discharge into a calyx.

SUMMARY

Two cases of hydatid cyst of the kidney are reported. In one, rupture and evacuation of the contents resulted in a spontaneous cure and in the other this was probably the case. In neither case was a positive early diagnosis reached in spite of suggestive evidence. In one case an unusual para-pelvic and extra-renal lodgement had occurred.

ACKNOWLEDGEMENT

Our thanks are due to Dr. Denis Stewart for his generous help in the pathology of these cases.

RUPTURE OF THE OESOPHAGUS

By M. P. Susman

Sydney

RUPTURE of the oesophagus may be due to:—

(1) Trauma, as from a surgical instrument, a foreign body or from external violence.

(2) Strain, as from vomiting or retching (Williams and Boyd, 1926; Loewenthal *et alii*, 1951; Foggitt, 1946), defaecation (Beal, 1949), childbirth, fits. This is the so-called spontaneous rupture.

(3) Disease of the oesophagus itself or of some adjacent organ or tissue.

Traumatic rupture is usually caused by a surgical instrument. It is more common and less serious than a rupture from strain mainly because it generally occurs in a person with an empty stomach who is already in hospital. The usual sites are the pharyngo-oesophageal junction and the lower end of the oesophagus. Strain rupture is almost always at the lower end and it generally occurs in a person with a full stomach who retches or vomits violently.

Rupture through diseased tissue usually has the added factor of trauma or strain and so overlaps the other two types.

In the past a number of persons with traumatic rupture of the oesophagus have recovered with conservative treatment and most people with strain rupture have died; because of this the differences between traumatic and strain rupture have been emphasized at the expense of the similarities although the two have, in fact, many problems in common. The results obtained from suturing the oesophagus are now better than they were (Jemerin, 1948), but so also are the results from conservative surgery. The common factor is improvement in the supporting treatment.

Barrett (1947) reported the first case of recovery from "spontaneous" rupture after suturing of the rent. He emphasized the bad results of conservative treatment as then practised; but comparisons between direct operation and conservative operation are valid only when both are adequate and associated with equally good management before and after the operation. Satisfactory conservative treatment would be especially valuable for old or feeble patients, or whenever adequate surgery and anaesthesia were lacking. Barrett's condemnation of conservative management of "spontaneous" rupture was too sweeping as he did not accept 4 cases of recovery after drainage of the secondary empyema. Since then 9 more cases of recovery on conservative treatment have been reported and there are at least 4 unreported cases (Table 1).

Moreover, nearly twenty years ago Gott (1933), giving a personal communication from P. P. Vinson as the reference, wrote "there are authentic cases of recovery, (from oesophageal rupture) without surgical interference." I have found reports of 9 recoveries after suturing of the oesophagus and add one of my own. It is therefore premature to say that suturing of the oesophageal rent is mandatory and there is no justification for Samson's (1951) assertion that the patient will die unless the rent is closed and that "so-called conservative drainage procedures have no place in the early hours following rupture." The same remarks apply to traumatic rupture of the oesophagus. Weisel and Raine (1952) report 7 cases of instrumental perforation successfully treated by immediate suturing; but this good record is well matched by the recoveries under conservative treatment reported by Adams (1946). Bisgard and Kerr (1949), although they advise closure of the oesophagus, remark that it usually re-opens. In Barrett's (1947) case the oesophageal rent was small, it had been closed with only a simple purse-string suture and it broke down during convalescence so that an oesophago-pleural fistula and empyema resulted. This sequence has occurred in other cases (Lynch, 1949; Temple, 1949) including mine with subsequent recovery of the patients.

TABLE 1

Cases of recovery from "spontaneous" rupture of the oesophagus treated conservatively.		Cases of recovery from "spontaneous" rupture of the oesophagus treated by suturing of the rent.	
1. Ballin and Salzstein	(1922)	1. Barrett	(1947)
2. Benson and Penberthy	(1938)	2. Olsen and Clagett	(1947)
3. Graham 4. Graham	(1944)	3. Scholefield	(1949)
5. Cummings, cited by Cliffton	(1946)	4. Temple	(1949)
6. Frink	(1947)	5. Lynch 6. Lynch	(1949)
7. Moore and Murphy	(1948)	7. Cliffton	(1949)
8. Cliffton	(1949)	8. Samson 9. Samson	(1951)
9. Lynch	(1949)	10. Susman	(1952)
10. Gardner	(1949)		
11. Ware and Streider	(1949)		
12. Vestal, cited by Cliffton	(1949)		
13. Dunavant and Skinner	(1951)		
14. Overholt, cited by Flipse	(1951)		
15. Tanner 16. Tanner personal letter	(1952)		
17. Kraeft and Hughes	(1952)		

One may well ask whether suturing of the rent is necessary and whether the added trauma and time justifies it. As the rent is almost always linear it will tend to close naturally if it is kept at rest and if it is kept free of contamination by means of constant gastric suction, which also saves the mediastinum and pleural cavities, from further soiling. I disagree with the opinion often expressed that gastric suction is not necessary because vomiting does not usually continue; but the contents of the stomach tend to pass into the oesophagus apart from vomiting, as in my Case 4. For this reason if parenteral feeding fails to maintain the patient, jejunostomy (Moore *et alii*, 1945) may be preferable to gastrostomy although the disadvantages of gastrostomy can be overcome by passing the tube through the pylorus into the duodenum (Goulston, 1952). Some writers (Healty, 1944; Kernan, 1950) have reported recovery from instrumental rupture with chemotherapy, antibiotics and supporting measures only, but because tension and infection in the mediastinum and pleural cavities are always serious, drainage of these areas and the prevention of further soiling are essential.

Study of some of the reported cases of rupture of the oesophagus are illuminating. For example, Lynch's (1949) first patient, a man of 45, was too ill for operation when first seen by the surgeon sixty hours after the instrumental rupture of the oesophagus. He had intercostal drainage, a late jejunostomy and general supporting treatment. Four attempts were made to close the rent and all failed. The fistula eventually healed and the patient recovered. Kraeft and Hughes' (1952) patient was a man of 55 whose oesophagus ruptured during a fit of coughing. He had to make a journey of 115 miles to reach hospital and he recovered after conservative management. His convalescence was interrupted by formation of a pericardial effusion and by empyema but the oesophageal fistula closed at last and in the end all was well. Kraeft and Hughes say

that this case shows "the fallacy and danger of conservative measures," but surely it rather shows that conservative measures may salvage a desperately ill patient who might well have died if more extensive surgery had been done.

Strong, Wilson and Taylor (1951) report 7 cases of rupture of the oesophagus; one was "spontaneous," one was caused by a foreign body and 5 were instrumental. The patient with the "spontaneous" rupture and 3 of those with instrumental rupture died. All were treated "conservatively." But we cannot condemn conservative treatment in favour of direct surgery on the strength of these results because in none of these patients was the stomach kept empty by suction and in none was the mediastinum or pleural cavity drained.

DIAGNOSIS

It is surprising how often an operator, after perforating an oesophagus with an instrument, is unaware of the damage he has done. The only safeguard is to assume that perforation has occurred if the procedure is followed by bleeding and increasing pain, especially if there are also any shock, cyanosis or dyspnoea. With strain rupture, there is a history of severe pain behind the sternum and in the epigastrium during some straining effort. Shock and fever soon follow and the diagnosis is confirmed by the finding of air in the subcutaneous tissues and mediastinal tissues and of air and/or fluid in one or both pleural cavities. The pleural effusion may contain gastric contents or methylene blue given by mouth; and iodized oil, given by mouth, may be shown in skiagrams to have passed into the mediastinum or pleural cavity.

TREATMENT

Prognosis depends partly on the time elapsing between the rupture of the oesophagus and the starting of treatment and the place for this is therefore the patient's home or the hospital casualty room: it should not be delayed until the patient is admitted to a ward. Whether it is decided to suture the rent or only to drain soiled areas, the following measures should be carried out as soon as possible:—

1. Tension pneumothorax should be immediately relieved by aspiration.
2. No food or fluid should be given by mouth.
3. Fluids should be given intravenously.
4. A tube should be passed into the stomach for constant suction. Its position should be checked by a skiagram; if it goes into the mediastinum or pleural cavity no harm is done and the diagnosis of rupture is further confirmed.
5. If a tube cannot be passed into the stomach a jejunostomy should be made, or a gastrostomy, with the tube passed into the duodenum.
6. Suitable antibiotics and drugs should be given.
7. Thoracotomy and mediastinotomy should be performed, with or without suturing of the oesophagus.

CASE RECORDS

I have to record 7 cases of rupture of the oesophagus. Five were strain or "spontaneous" ruptures. Three of these patients were moribund when first seen by me (Cases 1, 2 and 3); the fourth died seven weeks after the rupture (Case 4), and the fifth recovered after suturing of the rent (Case 5). Two of the ruptures were instrumental and both patients recovered with conservative management (Cases 6 and 7). All the patients came to treatment late.

Case 1

S.C., a man aged 75, had an attack of vomiting on the night before admission to hospital. He was then moribund and he died the next morning without a diagnosis having been made.

The important findings at autopsy were:—

(i) Both pleural cavities contained foul brown fluid and both lungs were oedematous.

(ii) The pericardium contained some turbid fluid and fibrin.

(iii) The mediastinum was generally but not grossly inflamed.

(iv) The oesophagus was normal except at the cardio-oesophageal junction; here there was a small extra-mural abscess around a small perforation. The mediastinal pleura on each side appeared to be intact.

Case 2

M.E., a women aged 64, had an attack of vomiting with pain in the lower part of the chest on the night before admission. Subcutaneous emphysema

of the neck was noted. Dark brown fluid aspirated from the right pleural cavity contained blood and alcohol. She died the same day.

The relevant autopsy findings were:—

(i) Subcutaneous emphysema of the neck and chest.

(ii) The mediastinum contained offensive fluid and the pleura on both sides was torn just above the diaphragm.

(iii) At this level there was a rent 1.5 cm. long in the oesophagus.

(iv) Each pleural cavity contained three pints of fluid and some food residue.

Case 3

T.W., a man aged 62, was admitted to hospital moribund, with the diagnosis of ruptured gastric ulcer. There was a history of vomiting followed by severe pain in the epigastrium and behind the sternum. Cyanosis and sweating were noted. He died the next day.

At autopsy:—

(i) There was a collection of milky fluid in the posterior mediastinum which bulged into both pleural cavities. A small hole was found in the mediastinal pleura on both sides.

(ii) Each pleural cavity contained similar milky fluid, and the pleurae were grey and opaque.

(iii) At the lower end of the oesophagus there was a tear 2 cm. long and the surrounding tissues were inflamed and irregular.

Case 4

J.O., a man aged 58, said that he had collapsed with severe pain in the epigastrium and back during a drinking bout the day before. The diagnosis of perforated gastric ulcer was made but the visiting surgeon thought that he was too ill for any operation. A tube was passed into his stomach and fluids were given by vein. The next day he was delirious, and clinical and radiological signs of right basal pneumonia were found. He did not improve with antibiotics and drugs. On the fourteenth day pus was aspirated from the right hemi-thorax. I did not see him until the following day, when thoracotomy for drainage was done; he remained ill and febrile in spite of adequate drainage. Three weeks later some of his swallowed fluids were found to be escaping through the empyema tube; it is probable that this had been occurring for some time before it was noticed. Feeding by mouth was stopped and a gastrostomy was made; fluid given through this promptly appeared in the drainage bottle. Parenteral fluids were given and the stomach was kept empty. Nine days after the gastrostomy was made he complained of pain and tenderness of both calves; thrombosis was diagnosed and heparin was given. Three hours later he was afflicted with precordial pain, dyspnoea, tachycardia, cyanosis and hypotension; an hour later he had a fit and died. This was forty-seven days after the rupture of the oesophagus.

At autopsy:—

(i) There was a large empyema cavity in the lower part of the right pleural cavity; it was lined by granulation tissue. The left pleural cavity was normal.

(ii) An abscess cavity in the right side of the mediastinum communicated with the empyema cavity.

(iii) Immediately above the diaphragm there was a hole in the right side of the oesophagus. There was very little inflammatory reaction present and a simple track went from the rent to the empty abscess cavity of the mediastinum and so to the empyema cavity.

(iv) No cause was found for the suddenness of his death.

Case 5

V.H., a woman aged 51, had swallowed caustic as a child following which she had had oesophageal dilatations for several years; but for most of her life she had had no dysphagia unless she swallowed hard or large substances. On the night of 6th April, 1950, a piece of meat stuck in her gullet: she tried to push it down by manual pressure at the root of the neck; this induced retching and almost at once she felt severe pain behind the sternum. Several hours later she was admitted to hospital in shock. Subcutaneous emphysema of the neck was noted by a house doctor but its significance was missed. She was worse next morning. Standard skiagrams of the lungs were normal, but swallowed barium was held up in the superior mediastinum and some of it passed out of the oesophagus. A tube could not be passed into the stomach; an intravenous drip was started. Her condition was very poor when I first saw her twenty hours after the onset of pain and a right thoracotomy through the 7th rib bed was performed soon afterwards. There was no air in the pleural cavity but a little turbid fluid was found when the lung was lifted forwards. There was a general bulging in the line of the oesophagus especially around the arch of the azygos vein. Some air and barium escaped when the mediastinum was entered. The mediastinal tissues were inflamed and manipulations around the oesophagus caused troublesome but not dangerous bleeding. Several silk sutures were used to close the rent which was about half an inch long at the level of the azygos arch; its relation to the stricture could not be made out. The chest was closed in layers after providing for two drainage tubes, one to the site of the perforation and one to the lowest part of the pleural space. She improved rapidly after the operation and the lung remained expanded and aerated; the higher of the two tubes was retained for eight days in case of leakage from the oesophagus. On the eleventh day a barium swallow entered the stomach without delay and irregularity of the upper part of the oesophagus was noted. On the way back to the ward she felt a sudden tearing pain in the chest and soon afterwards barium appeared at the opening of the upper tube track. The pain soon subsided, her condition remained good and at her own request she was soon taking a light diet which she swallowed without difficulty; the fistula and sinus track closed within the week, but three months

later pus came through the old sinus and clubbing of the fingers was obvious. She continued to swallow without difficulty and no food escaped. The sinus gradually closed and all was well until over two years later when a piece of meat became impacted in the upper end of the oesophagus and had to be removed through the oesophagoscope. She is now well again.

Comment: I think that the suturing of the oesophagus achieved nothing and should not have been attempted in the face of so much inflammation and oedema.

When it was found that a tube could not be passed into the stomach, a jejunostomy should have been made.

Feeding was begun too early, by mistake, and was then continued too liberally. Ounce doses of fluids were allowed within twelve hours of the operation and the quality and quantity were increased in the following days, mainly at the patient's insistence that she could swallow properly, which proved to be correct.

Despite these apparent errors in management the patient recovered.

Case 6

K.R., a woman aged 48, had had an oesophagoscopy fifty-two hours before I first saw her in consultation. She was very ill with extensive subcutaneous emphysema of the neck and face. There was no evidence of air or fluid in either pleural cavity. Cervical mediastinotomy was performed and air and turbid, flaky fluid escaped. A drainage tube was inserted and the wound was loosely sutured. She had nothing by mouth and was sustained by intravenous fluids. Antibiotics were prescribed. A week later a gastrostomy was performed by another surgeon. She gradually recovered and later a slight narrowing of the upper end of the oesophagus was found.

Case 7

D.B., a man aged 33, complained of attacks of vomiting and loss of weight. A barium examination showed delayed emptying of the lower end of the oesophagus and some dilatation above. Oesophagoscopy was done under general anaesthesia and five hours later cyanosis, dyspnoea and tachycardia occurred; there was little complaint of pain. Subcutaneous emphysema of the neck and a left hydropneumo-thorax were found. Two days later an oesophago-pleural fistula was shown in a radiogram after a swallow of iodized oil and also by the aspirating of swallowed methylene blue from the left pleural cavity. I first saw the patient at this stage and thought that he was too ill for the intended thoracotomy. The following treatment was given:—

(a) Nothing by mouth, constant gastric suction and intravenous fluids;

(b) closed intercostal drainage of the left pleural cavity;

(c) two million units of penicillin and 1 g. of streptomycin daily.

He improved rapidly on this treatment. Four days later better drainage of the chest was established through a rib bed and the lung soon expanded. Twelve days after the oesophagoscopy methylene blue by mouth did not appear in the drainage bottle and barium went straight into the stomach without any leakage. Feeding by mouth was then begun, the stomach tube was removed and the intravenous drip was discontinued. He was discharged three weeks later in good condition and was taking ordinary meals without difficulty. He has remained well.

SUMMARY

Rupture of the oesophagus is reviewed and 7 new cases are recorded. Surgery is a necessary part of the treatment and its scope is discussed. Particular attention is given to the merits of direct suturing of the oesophageal rent on the one hand, and of simple drainage of the mediastinum and pleural cavity on the other hand. The better results in recent years are due mainly to improvement in the supporting treatment; this must begin as soon as the diagnosis is even suspected.

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ADDENDUM

In the discussion that followed this paper a number of cases were reported by different speakers. Two new articles have lately come to hand; Anderson (1952) records four fresh cases of "spontaneous" rupture all of whom recovered after late suturing of the oesophagus (four or more days after the rupture). Anderson raises some doubt about the need for operation in these late cases; although he advises repair of the rent, he comments that if the patient survives 48 hours, conservative management offers good chances of recovery. He found that up to December, 1950, 92 cases of "spontaneous" rupture of the oesophagus had been reported, with 18 survivors; 12 new cases were reported in 1951, with 8 survivors. In the discussion of Anderson's paper additional cases were mentioned by various speakers, including 8 that recovered after a simple drainage operation.

Dugan (1952) deals with 16 cases of oesophageal perforation, 11 of which were due to endoscopic injury. He advises primary closure of tears of the lower end whether they are traumatic or "spontaneous."

Burcher (1952) is about to report a case of "spontaneous" rupture treated by suturing of the oesophagus and drainage of the pleural cavity. The suture line broke down on the twelfth day and closed itself about two months later. The patient, a woman aged 47, recovered.

Borrie (1952) has had a recent case. The rupture occurred in a middle aged woman during an attack of self-induced vomiting. At operation two days later a vertical tear in the lower end of the gullet was sutured and the left pleural cavity was drained. She made a good recovery.

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SUB-SEROSAL CYSTS OF THE PERITONEUM

By I. C. Heinz

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SURGEONS have often sent material to the pathologist for histological examination because, at laparotomy, small nodules scattered over the surface of the bowel and pelvic organs have presented a diagnostic problem.

The pathological processes which may give rise to such nodules are tuberculous peritonitis, carcinomatosis, talc granulomata, lycopodium peritonitis and multiple small cysts of the peritoneum. This paper is concerned with the cysts. They are commonly seen during operations or at autopsy but they receive little attention in the literature or in standard textbooks.

The following description of the condition is based on a series of 8 cases where biopsy material was examined and on observations made at other times when the cysts were seen *in situ*.

Clinical Features

The average age of the 8 cases was 37 years, the youngest being 25 and the eldest 51 years. There was no significant difference in the sex incidence. In all instances the cysts were an incidental finding during operation, or at autopsy where death was caused by some apparently unassociated condition. There had been no previous operation in these cases and a study of the clinical histories yielded no clue as to possible aetiological factors.

Pathological Features

The biopsy material sent for examination consisted of five appendices, two *uteri* and one Fallopian tube. No correlation seemed to exist between the cysts and other pathological changes present in this material and, in fact, they have more often than not been found on the surface of apparently normal organs.

Macroscopic appearance

The cysts are approximately 1 mm. in diameter and are almost invariably of uniform size. They are translucent and appear like grains of boiled sago scattered over the peritoneum where they usually occur in groups of two or three rather than singly.

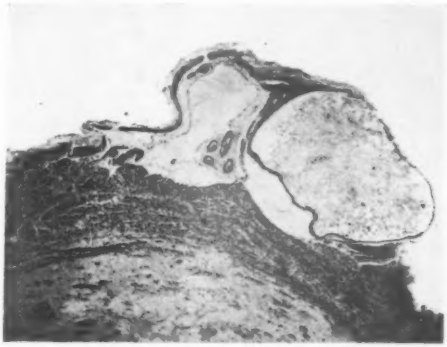

FIG. I. Photomicrograph of two cysts in the wall of an appendix. One is completely surrounded by fibres of the longitudinal muscle. (x 150)

The lesions have been observed on the Fallopian tubes, the posterior surface of the uterus and broad ligament and on the surface of the bowel as far up as the diaphragm. In some cases they are so numerous that there are few coils of bowel which do not present some cysts on their surface.

Histological appearance

Microscopically the cysts are seen to be lined by a single layer of flattened to cuboidal cells and they contain a mucinoid material with some amorphous debris and desquamated cells.

When, as is commonly the case, the cysts are embedded in the outer longitudinal muscle layer of the bowel then they also contain smooth muscle in their walls. Many

of the cysts, although projecting above the surface, are completely surrounded by smooth muscle (Fig. I). Other cysts are partly encircled by muscle and have a free surface composed of flattened cells which are quite distinct from the peritoneum covering the organ.

FIG. II. Photomicrograph showing a group of three cysts lying in the wall of an appendix. (x 150)

At what appears to be a later stage in the development of a cyst, most of the wall is extruded from the surface of the viscus, whilst fully formed cysts are simply thin-walled sacs with a narrow attachment to the peritoneal coat of the organ on which they are found (Fig. IV).

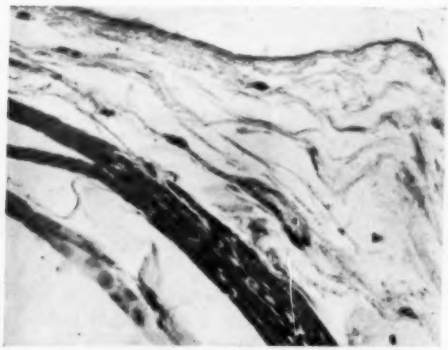

FIG. III. Photomicrograph of the superficial portion of a cyst wall showing the lining of the cyst on the left then the muscle and peritoneum. (x 350)

Many of the cysts lie alongside dilated lymphatic channels and morphologically it is impossible to distinguish between them. The association can be appreciated by observing the gradation between normal lymphatic vessels and fully developed cysts (Fig. V).

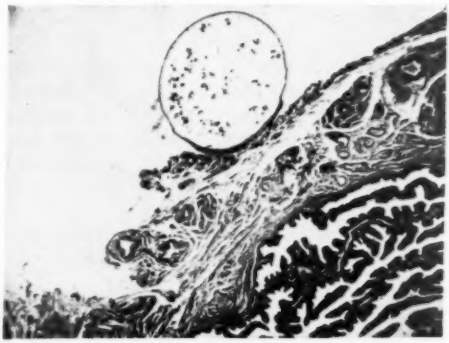

FIG. IV. Photomicrograph of a Fallopian tube with a more mature cyst attached to its wall. (x 50)

Although it is usual to find the cysts lined by a single layer of endothelial cells some show evidence of a proliferation of their lining membrane. In these circumstances many of the cells were cuboidal in shape with pale-staining cytoplasm (Fig. VI).

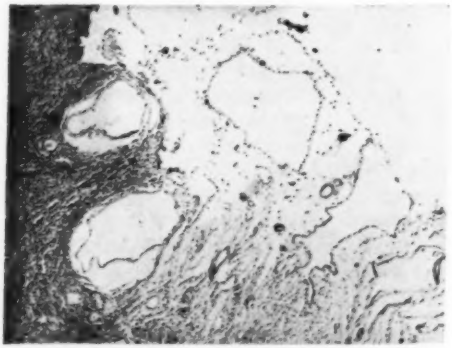

FIG. V. Photomicrograph showing the wall of an appendix with dilated lymphatics and fully developed cysts. (x 150)

Frequently one sections what appears to be a small cyst only to find that it is a nodule composed of a solid mass of rounded. vacuolated cells. These nodules apparently have their origin in a proliferation of the

serosal cells and the resulting area of hyperplasia may either lie embedded in the wall of the viscus or else be pedunculated (Figs. VII and VIII). Some of the nodules show central cavities of varying sizes. A study of these demonstrates that cysts may also form following a central necrosis of the cells forming these solid nodules.

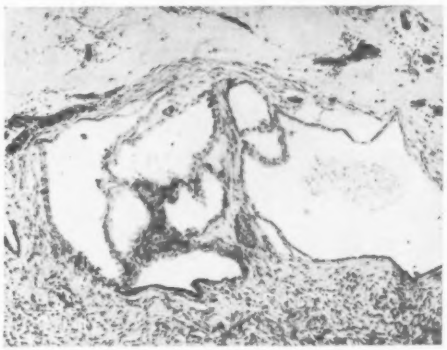

FIG. VI. Photomicrograph of cysts in the wall of an appendix showing proliferation of their lining cells. (x 150)

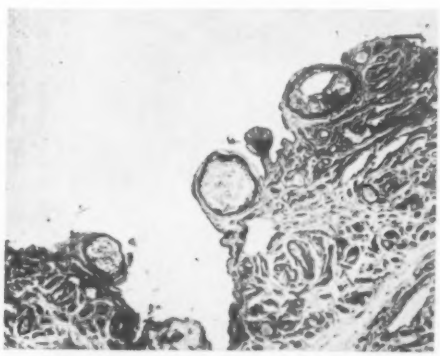

FIG. VII. Photomicrograph of a group of developing cysts on the Fallopian tube. There is central cavitation in one nodule. (x 50)

DIAGNOSIS

In many cases where these cysts are seen at laparotomy their minute size and small numbers indicate that they are of no pathological significance. Sometimes, however, when the cysts are numerous they are confused with tuberculous peritonitis, but the uniform size of the nodule should enable a diagnosis to be made in most cases. In the same way their minuteness and regularity usually serves to differentiate them from carcinomatosis, talc granulomata or lycopodium peritonitis. The histological appearance of these cysts is, of course, quite characteristic.

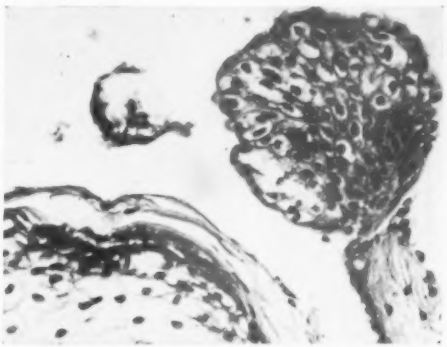

FIG. VIII. Photomicrograph showing the pedunculated nodule seen in Fig. VII. (x 400)

DISCUSSION

Multiple small cysts of the peritoneum are a definite pathological entity and the designation, sub-serosal cysts of the peritoneum, seems to be an appropriate name for them. These minute cysts have often been described on the ovaries, the Fallopian tubes and the broad ligaments. There have been many theories advanced to explain their occurrence in these situations but no fundamental difference appears to exist between this group and those occurring in the general peritoneal cavity.

A study of the above material indicates that sub-serosal cysts may form in two ways. Firstly by a dilatation of lymphatic channels lying either in the external muscle coat of the bowel or in the sub-serosal areolar tissue of the bowel and other organs. Secondly, they may have their genesis in a localized proliferation of serosal cells, the hyperplastic nodules so formed then breaking down to form the typical cysts. Although

both methods of formation can be seen on the female pelvic organs only the first method was observed in the general peritoneal cavity.

In both these types the aetiological factors are obscure.

SUMMARY

A pathological entity consisting of multiple small cysts of the peritoneum is described and it is suggested that they be called subserosal cysts of the peritoneum. Their origin from lymphatics and from a proliferation of the serosal cells is discussed. The cysts are of no clinical significance but sometimes need to be differentiated from other pathological conditions seen in the peritoneal cavity.

ACKNOWLEDGEMENT

I would like to express my gratitude to Professor E. S. J. King for giving me access to his Jacksonian Prize essay for 1930, "The Pathology of Ovarian Cysts." This was helpful both in the material it contained and in providing references to the literature.

A CASE OF DOUBLE UTERUS WITH SEPTATE VAGINA

By A. Eisdell Moore
Auckland

ABNORMALITIES in the female genital tract are by no means uncommon. Jean Burton-Brown (1948), writing from the Nuffield Department of Obstetrics and Gynaecology in the University of Oxford, says, "Defects of the genital tract are not always obvious, and many women pass through their confinement without the midwife or doctor becoming aware of their presence. Even when the defect involves the external organs many women reach adult life without suspecting that there is anything unusual in their anatomy."

This naturally makes it very difficult for anyone accurately to estimate the incidence of these defects. Smith (1931) recorded a rate of reported abnormalities of 1 in 7,040 cases in general hospital records, and compared this with a rate of 1 in 1,438 cases, when for five and one-half years he made a special examination for these defects in all his hospital admissions. As some of these cases, although symptomless in themselves, are associated with urinary abnormalities for which hospital treatment has been necessary, it is probable that the frequency of the real symptomless uterine deformity lies somewhere between these two figures, perhaps 1 in every 3,000 females.

Abnormalities depend upon:—

(a) Failure of the Müllerian ducts to develop, resulting in absence of uterus with or without absence of the vagina. This deformity is not likely to be overlooked.

(b) Failure of the Müllerian ducts to fuse, resulting in varying degrees of bicornuate uterus.

(c) Persistence of the medial septum, either complete with double vagina, or incomplete with sub-septate uterus or septate vagina.

For practical purposes this means,

(i) that the uterus may have a dividing septum of varying depth;

(ii) that the uterus may have two bodies with common cervix;

(iii) that two separate uteri may be fused together, each with its own *os* opening into a common vagina.

The deformities resulting from any of these varieties can easily be overlooked, as normal menstruation, normal sexual intercourse, and normal pregnancy may occur on one or both sides.

Dysmenorrhoea, menorrhagia, or other menstrual irregularities may occur, but it is recognized that in the *uterus duplex* abnormality menstruation may take place first from one organ and then from the other.

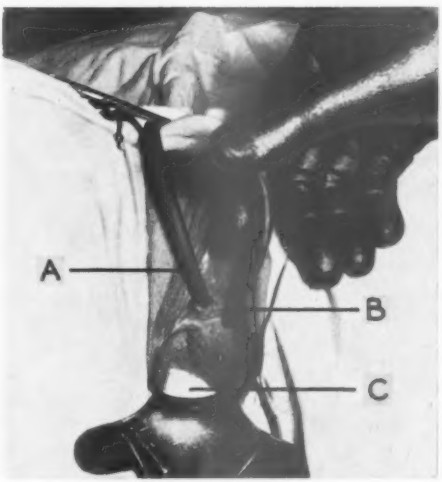

FIG. I. (A) Catheter in the urethra.
(B) Dilator in the sinus.
(C) Packing in the vagina.

Case History

Miss J.F. was referred to me on 2nd June, 1949, because of a very thick resilient hymen. Advice had been sought for routine pre-marital overhaul, and the patient was symptomless apart from some dysmenorrhoea. From the menarche her periods had always been heavy, and often as long as ten

days, but only occasionally with sufficient pain to keep her away from school or from her work. Recently the pain had become more severe.

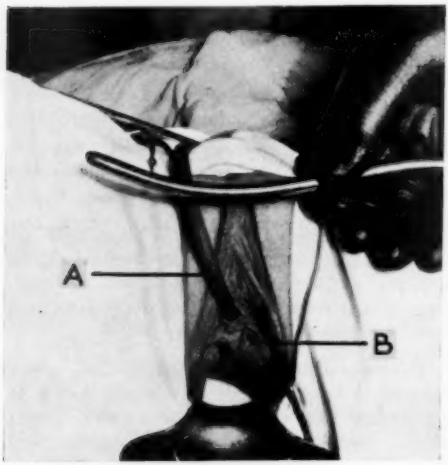

FIG. II. Showing the length of the dilator introduced.
(A) Catheter in the urethra.
(B) Opening of the apparent sinus.

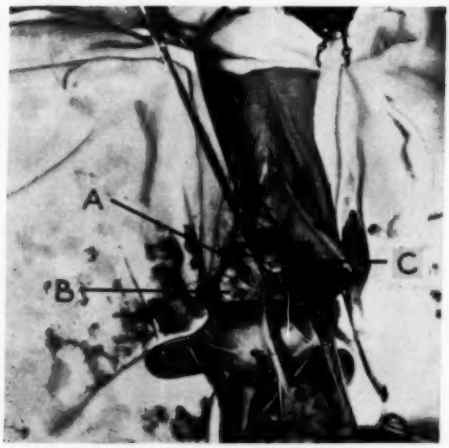

FIG. III. Showing the septum and packing in the separate vaginae.
(A) Septum.
(B) Gauze in the right vagina.
(C) Gauze in the left vagina.

Hymenectomy was performed on 13th June. The hymen was found to be very thick and rigid, with a central opening of about ¼ inch in diameter. A smaller opening about ⅛ inch in diameter half an inch to the left of the urethral orifice was noticed just inside the left *labium minor*, and from this a few drops of mucus could be obtained. A probe was passed in some distance, and the opening was thought to be that of a sinus leading from a cyst of the vaginal wall.

The patient was subsequently told of this sinus, but refused further investigation.

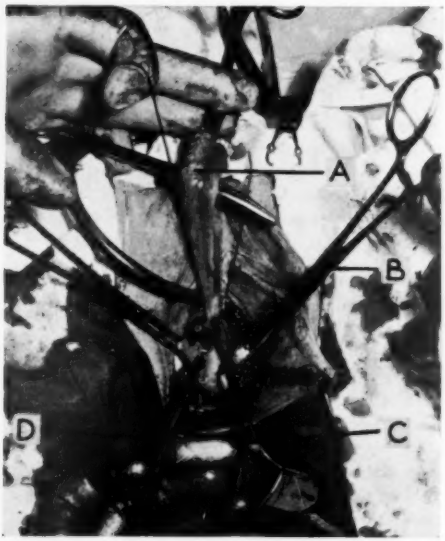

FIG. IV. The cervix is pulled down to introitus and septum pulled forward.
(A) Separated septum still attached to cervix.
(B) Forceps dragging the cervix down.
(C) Dilator placed in left *os*.
(D) Dilator placed in right *os*.

Three years later, now as Mrs. A.H., she came to see me on 16th July, 1952, saying that she had been married for two years, had had a normal pregnancy and confinement, and that her baby was now a year old. Throughout the pregnancy she had been worried by a continuous discharge of mucus from the sinus, necessitating constant diaper protection, and this discharge was still present.

Examination showed the opening to be a little larger than previously, and the smallest uterine dilator when passed was felt to reach the cervix. The vagina through which a child had recently passed was quite capacious, and the sinus apparently ran up along the left vaginal wall.

A tentative diagnosis was made of a Nabothian cyst of the cervix bursting through into the tissue of the vaginal wall and then tracking down to the introitus, and excision was advised. As the condition appeared to be unusual, arrangements were made for a photograph to be taken at the operation.

Fig. I shows appearance at operation with a catheter in the urethra and a small uterine dilator in the sinus.

Fig. II indicates that the dilator had been passed to the level of the cervix.

On starting to dissect away the track it was found that the apparent mouth of the sinus was in reality a small opening in a hymen which practically occluded a full-sized collapsed second vagina, into which a normal but somewhat small cervix protruded.

Fig. III shows the double vagina with gauze packing on each side of a very thick septum.

FIG. V. X-ray showing associated absence or abnormality of the left kidney.

The inter-vaginal septum was cut away, and Fig. IV shows it held forward just before complete severance from the cervix. A uterine dilator has been introduced into each *os*. Both uteri accepted dilators for about 4 inches and were apparently quite separate except for the fused cervix.

As the patient's only complaint had been the mucous discharge it was thought unjustifiable to perform a laparotomy to verify these findings.

The practitioner who attended her during her recent confinement confirmed the history of continuous mucus discharge throughout the pregnancy which was normal in other respects, and stated also that the labour was natural except for some delay in the third stage necessitating a small episiotomy.

As these cases are frequently associated with urinary abnormalities, an intravenous pyelogram was done, the report of which is as follows:—

> "No excretory shadow is seen from the left kidney. Excretion in good concentration is shown from the right kidney. The kidney pelvis and ureter appear wide but normal cupping of the calyces is shown. The bladder shadow appears normal. The widening of the pelvis and ureter may be due to compensatory hypertrophy." (Fig. V.)

The patient had never had any urinary symptoms.

SUMMARY

1. The frequency of abnormalities of the female genital tract is discussed.

2. A case is described where the persistence of a mucus discharge at the vulva led to the discovery of a second vagina with accompanying *uterus didelphys*.

3. Intravenous pyelogram showed associated urinary abnormality.

ACKNOWLEDGEMENT

I am grateful to Mr. Litherland, clinical photographer to the Auckland Hospital, for taking photographs illustrating this case.

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MYOMA OF THE STOMACH

A REVIEW OF 50 CASES

By T. Antonie

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THE incidence of smooth muscle tumours arising in the stomach is low, and in the clinical practice of most surgeons their infrequent appearance has caused them to be regarded by many as a rarity. Nevertheless, a statistical survey of most of the larger clinics reveals a significant number of cases. It is perhaps not generally realized that gastric myomata, benign and malignant, are second only to carcinoma in order of occurrence.

Of the benign neoplasms, the innocent smooth muscle tumour is the commonest, of which it constitutes 36.6 per cent. Timonem (1948), using a clinical rather than a pathological classification, gives the following analysis:

Myomata	36.6 per cent.
Fibromata	19.3 per cent.
Polypi	19.2 per cent.
Neurofibromata	10.9 per cent.
Papillomata	9.5 per cent.
Adenomata	4.5 per cent.

Considering the malignant members of the group, Marvin and Walters (1948) found at the Mayo Clinic one myosarcoma to 625 cases of carcinoma.

The present series is derived from the records of the Pathology Departments of the Royal Melbourne and Alfred Hospitals and covers a twenty-seven year period from 1925 to 1951. In all, 50 cases were reviewed, a few being referred from private practice for opinion.

General Characteristics

Presenting Features

In this series more than half of these tumours were symptomless, being detected accidentally, either at laparotomy for some unconnected condition, or, more commonly, during routine examination at post-mortem examination.

Table 1

SUMMARY OF FINDINGS

Period:	27 years
Number of cases:	50
Sex incidence:	
Male	22
Female	28
Age incidence:	
Youngest (Case 18)	22 years
Oldest (Case 4)	87 years
Chief Method of Presentation:	
Found accidentally at post-mortem	22
Found accidentally at operation	6
Haematemesis and melaena	14
Dyspeptic symptoms and pain	8
Abdominal tumour	3
Incidence of Malignancy:	
Malignant	4
Doubtful innocence	8
Multiple tumours:	5

Fourteen cases, almost one quarter, presented with haematemesis and melaena usually preceded by mild dyspepsia and epigastric discomfort. In the majority of these blood loss was severe. There were three deaths—one, an old woman of 87 (Case 4, Fig. III), died from the direct effects of haemorrhage and two others (Cases 31 and 37) succumbed in the immediate post-operative period.

The youngest member of the series, aged 22, was admitted with haemoglobin estimation of 22 per cent. She was submitted to laparotomy and the local removal of an ulcerating tumour of the lesser curvature.

In Case 28, a man of 72 years suffered severe haematemesis and melaena unresponsive to repeated transfusion over three days. The clinical picture resembled that of a bleeding peptic ulcer. Laparotomy revealed ulceration of a fibromyoma.

TABLE 2

AGE DISTRIBUTION OF 50 CASES ACCORDING TO DECADES

	20–30	30–40	40–50	50–60	60–70	70–80	80–90
				52			
				52			
				52			
				53			
				53			
				53	60		
				55	60		
				55	61	70	
				55	61	70	
				56	62	72	
			43	57	63	73	
			44	57	63	73	
			45	58	65	73	80
		32	45	58	66	75	83
		37	45	59	68	76	84
	22	39	47	59	69	79	87
Total	1	3	6	16	11	9	4

Eight patients sought advice for dyspeptic symptoms and pain. Most of those presenting with haemorrhage, admitted to previous indigestion and epigastric discomfort. Carcinoma was naturally suspected in many of these instances. Four of these eight growths were actively growing sarcomatous tumours. The fifth (Case 23), although benign, produced its symptoms by virtue of its juxtapyloric situation.

A painless upper abdominal swelling was the initial presenting feature in one case (Case 25),—the barium meal showed a filling defect on the greater curvature. The tumour had invaded the gastro-colic omentum and was histologically active. Death occurred two and a half years later — presumably from metastases. In two others (Cases 26 and 30) an epigastric mass was detected at the first examination.

Necrotic change leading to perforation and peritonitis has been recorded but was not found in the present series.

Elgood (1951) reports a case of gastro-duodenal intussusception caused by a large leiomyoma of the stomach.

Macroscopic Appearances

Here there is considerable variation, many of the lesions being small and not productive of symptoms. In four cases there were two tumours. In Case 43 a small group of nodules was found, the largest of which had a diameter of one centimetre. They may arise in the sub-peritoneal, sub-mucous or interstitial layers (Figs. I and II).

FIG. I. Photograph of the stomach from Case 6 showing two discrete tumours lying in the sub-peritoneal layer towards the fundus.

Tumours from the inner layers grow centrally towards the lumen where there is little resistance to their expansion. Generally they are small as in Cases 1 and 6. Myomata from the external layers grow outwards, often become pedunculated and may be quite large. Case 25 lends support to this last suggestion.

In this series there seems to be no special predilection for any part of the stomach. Distal and proximal parts are equally affected, whilst both curvatures would appear to be equally involved.

As would be expected the larger tumours are prone to areas of haemorrhage and cystic change. This is well seen in Case 4 (Fig. III). Central cavitation may communicate with the exterior, that is, with the cavity of the stomach as in Case 9.

Ulceration was noted in many of the submucous variety, being responsible for chronic or even massive blood loss, although it is of interest that haemorrhage may occur

without actual demonstrable breach of surface continuity, as in Case 4 (Fig. III). The remarks of Timonem (1948) are of interest in this regard. He believes that severe bleeding is likely to occur in the narrow muscular confines, for example, the pylorus—according to the *caput succedaneum* theory, that is, the projecting tumour becomes subjected to a ring-like pressure and consequently fluid circulation is impeded and stasis, oedema and even haemorrhage arise below the ring.

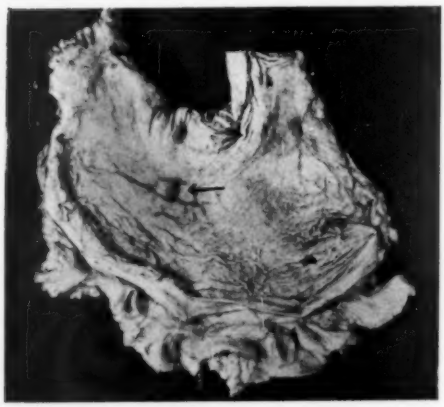

FIG. II. A photograph from Case 1, showing the mucosal surface of the stomach. A single myoma is seen on the posterior surface in the sub-mucosa, midway between cardia and pylorus.

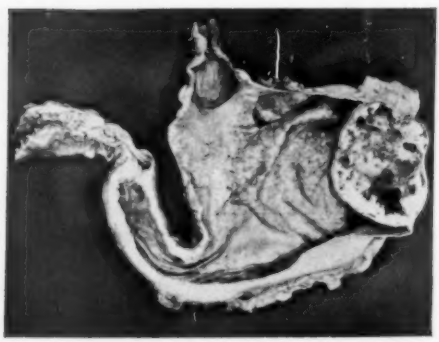

FIG. III. Specimen removed at autopsy from Case 4, showing an encapsulated tumour 6.5 cm. in diameter on the greater curvature. Areas of haemorrhage and cyst formation are seen.

Lobulation may be a feature but, as a rule, the lesion is single, encapsulated and well demarcated. A frankly invasive character clearly indicates malignancy as in Case 19 in which there was direct extension into the pancreas.

Cole and Barry (1950) emphasize that even in well-circumscribed lesions and in the absence of obvious metastases or invasion of surrounding organs by tumour, it is impossible to determine by the gross appearance whether the tumour is benign or malignant, as in Case 15 of this series.

Myomata are occasionally found to co-exist with other commonly occurring gastric lesions. In Case 8 there was a concomitant carcinoma—and in Cases 21, 38, 40 and 41 a chronic peptic ulcer.

FIG. IV. Low power photomicrograph of portion of the wall of stomach including the tumour from Case 17. The mass of muscle tissue separated from the mucosa in this area by a somewhat oedematous sub-mucosa is clearly seen. (x8)

Microscopic Appearances

Histologically the picture is variable. The majority of tumours recorded here are benign, composed of adult muscle cells containing elongated blunt-ended nuclei as

shown in Fig. V. Fine whorling or palisading is a common arrangement. The gross appearance of encapsulation may be confirmed microscopically (Fig. IV).

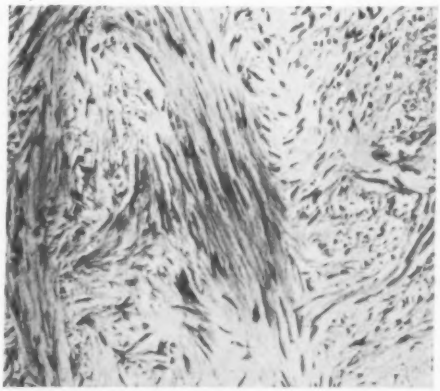

FIG. V. Photomicrograph of portion of the tumour from Case 20. The whorled strands of muscle tissue are well demonstrated and the characteristic elongate nuclei with blunt ends are to be seen. (x 150)

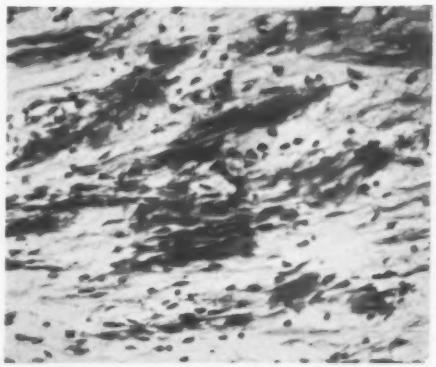

FIG. VI. Photomicrograph of portion of the tumour from Case 23. Groups of muscle cells lie in a fibrous stroma, the tumour thus being a fibromyoma. (x 150)

In many instances the tumours are relatively avascular and quiescent. Adult fibrocytes may be recognized by the fusiform outline of the nuclei and adjacent strands of collagen and these may be intimately mixed with muscle cells. Special stains, for example Van Giesen or Masson, are sometimes required to demonstrate the exact proportion of each. These tumours are fibromyomata and microscopically resemble those occurring in the uterus. They exhibit the same liability to hyaline change and calcification (Fig. VI). Local areas of necrosis with cyst formation and haemorrhage are frequently seen as in Case 4.

FIG. VII. Photomicrograph of portion of the tumour from Case 18. This tumour is much more cellular than those shown in Figs. V and VI, but the nuclei are of the narrow elongate adult form. (x 150)

All gradations of cell pattern, vascularity and oedema are encountered, with variability of stromal content. In some there is extreme cellularity with little stroma, the cells being atypical — often rounded or irregular (Figs. VII, VIII and IX).

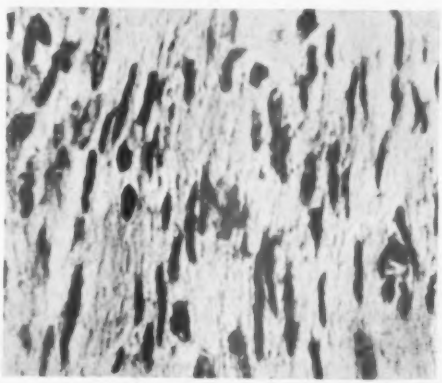

FIG. VIII. Photomicrograph showing the characteristic adult form of the cells shown in Fig. VII. (x 700)

All grades of activity are seen. Uniform arrangement may be lost, the cells running in a disorganized way which is frankly anaplastic and malignant. Confirmation is given by the dark-staining nuclei and occasional mitotic figures (Fig. X).

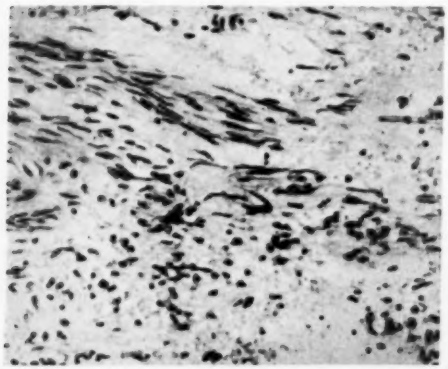

FIG. IX. Photomicrograph of portion of the tumour from Case 27. The strands of muscle tissue have a typical form and some necrosis, which is a prominent feature in some of these tumours, is shown at the right and lower part of the photograph. (x 150)

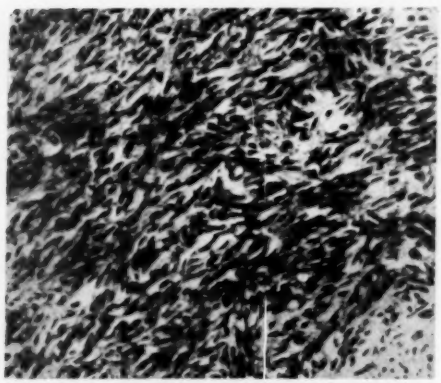

FIG. X. Photomicrograph of portion of the tumour from Case 19. The tissue is very cellular and the cells are irregularly arranged and contain large active nuclei. The degree of activity is such as to justify the diagnosis of malignant myoma. (x 150)

A categoric diagnosis of malignancy on histological grounds alone is often difficult as in Case 9. Of itself, cellular activity is not diagnostic, nor indeed is the presence of an isolated mitotic nucleus (Cases 33 and 41). On the other hand, a metastasizing leiomyoma with gross secondary deposits in the liver or lungs is occasionally encountered where the histological appearances suggest innocence. Marvin (1948) reported lymph node involvement in two cases.

DIAGNOSIS

(a) Radiological:

Accurate diagnosis of myoma, or at least of a benign tumour, is often possible by barium meal examination. Chronic dyspepsia or even repeated haemorrhage, presumed for years to be due to an ulcer, may be shown on such investigations to result from a gastric myoma.

FIG. XI. An X-ray photograph from Case 24. The large filling defect in the proximal part of the stomach has the appearances of an innocent tumour. Histologically it was an actively growing myoma.

A filling defect is, as a rule, observed and may, of course, be mistaken for carcinoma. However, on careful screening, uninterrupted peristalsis often indicates the absence of infiltration and the correct diagnosis may be suspected even if not confirmd. Fig. XI (Case 24) illustrates the typical appearances of a smooth muscle tumour.

In this series 15 cases were examined radiologically—in 5 the correct diagnosis was suggested (Cases 18, 26, 30, 32 and 34); in 5 carcinoma was suspected (Cases 10, 29, 33, 35 and 36) and in others a non-committal descriptive report was given.

Case 17, suspected of being an extra-gastric lesion, was, in fact, a pedunculated sub-peritoneal tumour.

Calcification, if seen, supports the diagnosis of innocence.

Husebye (1948) emphasizes the significance of air-filling of the stomach when doubt exists. A perfectly smooth, round, regular, barium-outlined contour is a feature of the innocent rather than malignant tumour, as in Case 18 (Fig. XII).

FIG. XII. An X-ray photograph from Case 18, showing a semi-circular filling defect in the distal porion of the lesser curvature. The smooth and rounded contour is characteristic of the encapsulated myoma.

(b) Gastroscopic:

Where doubt exists following radiological examination, gastroscopy may help in the elucidation of the problem. In Case 32 this investigation confirmed the innocent nature of the tumour demonstrated radiologically.

(c) Clinico-Pathological:

Fractional test meal is of little diagnostic significance but in Case 10 where carcinoma was suspected radiologically, and blood and mucus were found in all specimens of gastric contents, the presence of free and total acid in normal amounts rendered the original diagnosis suspect.

(d) Differential Diagnosis:

(i) On clinical considerations symptoms of chronic pain and dyspepsia will require elucidation to differentiate the more commonly encountered peptic ulcer or carcinoma. A careful history of the pain, taking into account its type and time relationships, its periodicity or chronicity, its aggravation or relief, its progressive or static nature, will often allow the correct diagnosis to be suspected and later confirmed.

Haemorrhage and more rarely perforation are difficult to differentiate on clinical grounds alone.

(ii) On radiological appearances smooth muscle tumours of the extra-gastric variety usually have a smooth, rounded outline causing little interference with adjacent peristaltic movement. Nevertheless, confusion with a fungating carcinoma may arise.

When ulceration is gross, peptic or neoplastic conditions are naturally to be suspected.

A pedunculated tumour arising from the outer coats of the stomach may cause indentation of the barium-outlined viscus, giving the erroneous impression of an extra-gastric tumour as in Case 17.

(iii) On the pathological considerations other benign tumours, especially neurofibromata, resemble the smooth muscle tumours not only clinically and radiologically but also sometimes in their macroscopic and microscopic appearances. Experience with special staining methods may be required in these instances.

TREATMENT

The principles governing treatment depend fundamentally on the nature of the tumour —benign or malignant. From the foregoing it has been shown that a distinction cannot always be made easily or indeed with certainty.

It is true that half of the cases in this series were diagnosed by accident at post-mortem and were obviously innocent. It is equally true that 4 cases were frankly sarcomatous but 8 cases, at least, were regarded by the pathologist with suspicion and doubt. Malignancy was suggested by the use of the term "actively growing," but a frank designation was withheld.

Where doubt exists, as it often does, the operating surgeon must assume malignancy and act accordingly. Not infrequently the malignancy is low and radical resection is rewarded by long survival.

Two other factors, local recurrence and lymph node involvement, both of which are occasionally encountered, are again sound arguments for the radical approach.

Whilst not seen in this series, instances of local recurrence have been noted. Dixon and Kratzer (1951) report leiomyosarcoma occurring in the stump of the stomach many years after resection for leiomyoma. In their case a Polya gastrectomy was performed on a child of nine for a leiomyoma. Twenty-four years later, dyspepsia and weight loss were reported, barium meal showing a lesion in the gastric stump. Laparotomy revealed a lobulated, ulcerated, infected lesion, requiring further gastric resection. The ulcer had perforated and involved the liver, and microscopically was sarcomatous. Recurrence was assumed without excluding the possibility of multicentric tumour formation.

Lymph node involvement by non-epithelial tumours is unusual. Nevertheless, it may occur with smooth muscle as well as with other sarcomata.

From these considerations the inference must be for wide initial excision of the tumour with an adequate portion of healthy stomach wall and the regional lymphatic field.

However, from the practical viewpoint it should be realized that many, and perhaps most, of these tumours are innocent. Often they arise in inaccessible situations, for example the fundus, and not infrequently in the aged and infirm, in whom radical surgical procedures will result in a high mortality. Local removal will no doubt be reasonable under such circumstances.

Nevertheless, the surgical requirements based on the pathology of these tumours as a whole, will only be fully satisfied by a radical ablation.

CONCLUSIONS

1. Fifty cases of smooth muscle tumours of the stomach are reviewed.

2. More than half were without symptoms, being found accidentally. Dyspeptic manifestations were common. Acute or chronic blood loss was seen in almost a quarter of the cases. Three patients presented with an abdominal swelling. Perforation has been reported.

3. Any part of the stomach and any of its muscular layers may be the site of origin. The tumours are variable in size and number but generally they are single and small. Necrotic change with ulceration, cavitation and haemorrhage were seen. A growth cannot be designated benign or malignant with any certainty from the gross appearances.

4. The microscopic picture is variable in cell pattern, vascularity and stromal content. When malignancy is suspected clinical behaviour, rather than cellular morphology, is often a more reliable index.

5. When symptoms exist the nature of the lesion may be suspected on clinical grounds but radiological and gastroscopic studies should lead to the correct diagnosis in a high percentage of cases.

6. Distinction from other benign neoplasms is, as a rule, impossible without histological examination and even then may present difficulties. Malignant myomata are mistaken for carcinoma whilst occasionally an extra-gastric tumour leads to confusion.

7. Individual circumstance must always be an important factor in determining the correct surgical treatment in any case, nevertheless a radical removal is advised where practicable.

TABLE 3

A SUMMARY OF DETAILS OF THE FIFTY CASES REVIEWED

This table is derived from a study of clinical records. It shows the incidence and descriptive features of the smooth muscle tumours, together with their mode of presentation, course and, where available, their treatment and end result.

Case	*Age*	*Sex*	*Number*	*Size (diam.)*	*Method of Presentation*	*Macroscopic Description*	*Radiological Features*	*Histology*	*Clinical Course, Treatment, End Result*
1 (G.H.)	55	M	1	1.5 cm.	P.M.	Sub-mucosal. Midway between cardia and pylorus	—	Myoma—benign	Symptomless. Death from other causes
2 (F.P.)	83	F	2	7 mm.	P.M.	Sub-peritoneal	—	Myoma—benign	Symptomless. Death from other causes
3 (L.C.)	57	M	2	"Small"	P.M.	Sub-serosal at fundus	—	Encapsulated myoma	Symptomless. Death from other causes
4 (H.C.)	87	F	1	6.5 cm.	Haematemesis and melaena	Greater curvature	—	Myoma. No ulceration—areas of haemorrhage	Death from haematemesis and melaena
5 (L.W.)	73	F	1	"Fairly large"	P.M.	Sub-mucosal. Midway along lesser curvature	—	Myoma—benign	Symptomless. Death from other causes
6 (R.W.)	84	M	2	7.5 mm.	P.M.	Sub-peritoneal. At fundus	—	Myoma—benign	Symptomless. Death from other causes
7 (W.K.)	62	M	1	"Small"	P.M.	Sub-mucosal. On anterior wall of greater curvature	—	Oedematous submucous myoma	Symptomless. Death from other causes
8 (S.McF.)	69	F	1	"Small"	P.M.	Sub-peritoneal. At fundus	—	Fibromyoma	Death from cardiac failure. P.M. revealed a carcinoma of the pylorus, also a small fibromyoma at the fundus
9 (E.D.)	39	F	1	—	Haematemesis	Fleshy. Two cavities communicating with lumen	—	Myoma. Spindle cells; probably innocent	Dyspepsia culminating in haematemesis. Laparotomy. Local removal
10 (A.E.)	60	F	1	1.5 cm.	Haematemesis and melaena	Sub-mucosal with ulceration; on greater curvature near pylorus	Filling defect regarded as carcinoma	Myoma—benign	Anaemia for 4 years. Dyspepsia for 2 years. Fractional test meal showed blood and mucus in all specimens but normal acid. Laparotomy. Local removal
11 (A.B.)	58	F	1	"Small"	P.M.	Sub-mucosal at fundus	—	Myoma—benign	Symptomless. Death from other causes
12 (W.M.)	45	M	1	"Small"	P.M.	Sub-mucosal at fundus	—	Myoma—benign	Symptomless. Death from other causes
13 (F.B.)	69	M	1	"Small"	P.M.	Sub-mucosal. On anterior wall near cardia	—	Myoma—benign. Areas of degeneration and calcification	Symptomless. Death from other causes
14 (G.T.)	70	M	2	"Small"	P.M.	Sub-mucosal near cardia	—	Myoma—benign	Symptomless. Death from other causes
15 (S.H.)	47	M	1	"Large"	Epigastric pain and dyspepsia	Mobile, sub-mucosal, encapsulated, on posterior wall near pylorus	—	Degenerating myoma. ?Sarcoma	Four months indigestion, loss of weight, epigastric pain. Laparotomy and local removal
16 (J.T.)	58	M	1	"Small"	P.M.	Sub-mucosal at cardia	—	Myoma—benign	Symptomless. Death from other causes
17 (?.B.)	66	F	1	15 cm. 1 kg. weight	Haematemesis	Sub-serosal. From lesser curve into lesser sac. Pedunculated, multilocular	Large filling defect regarded as due to extrinsic pressure	Leiomyoma with degeneration	Laparotomy. Sleeve resection of stomach and tumour. Alive and well two years later
18 (M.L.)	22	F	1	3.7 cm.	Haematemesis and melaena	Sub-mucosal, pedunculated. Some ulceration	Circular filling defect. ? Myoma ? Lipoma	Fibromyoma	Laparotomy. Removal by submucous dissection

TABLE 3 — (continued)

Case	*Age*	*Sex*	*Number*	*Size (diam.)*	*Method of Presentation*	*Macroscopic Description*	*Radiological Features*	*Histology*	*Clinical Course, Treatment, End Result*
19 (F.F.)	68	M	1	"Large"	Dyspepsia	Fungating tumour in fundus invading pancreas	—	Malignant	Dyspeptic pain 1 year. Excision of fundus of stomach with growth and oesophago—gastrostomy. Died 1 year later with liver metastases
20 (E.T.)	63	F	1	"Small"	Found accidentally at operation	Nodule on lesser curvature	—	Myoma—benign	Operation for cholecystectomy. Nodule on lesser curvature removed
21 (A.S.)	55	M	1	2 cm.	Found accidentally at operation	Sub-serosal nodule	—	Spindle cells. Areas of hyaline change and calcification	Laparotomy for penetrating pyloric peptic ulcer—sub-serosal myoma also removed
22 (C.H.)	32	F	1	—	Dyspepsia	Pyloric tumour	—	Very cellular. Sarcoma	Seven months pain and dyspepsia. Local, transgastric removal of tumour
23 (V.W.)	52	F	1	"Small"	Dyspepsia	Hard nodule on posterior wall of pyloric antrum	—	Fibromyoma	Six months pain and dyspepsia. Hard nodule found. Partial gastrectomy
24 (M.F.)	32	F	1	6 cm.	Haematemesis	Sub-mucosal at fundus	Large filling defect on lesser curvature near cardia	Myoma—actively growing	Pain, dyspepsia, anaemia, culminating in haematemesis. Regarded as carcinoma. Laparotomy. Submucous removal of tumour
25 (? P.)	61	M	1	10 cm.	Abdominal tumour	Cystic tumour in gastrocolic omentum	Filling defect	Myoma—actively growing	Three months painless epigastric mass. Local removal. Patient died 2½ years later. ? Cause
26 (? W.)	70	F	1	5 x 4 x 2.5 cm.	Abdominal pain. Epigastric mass	Partly solid, partly cystic. Areas of haemorrhage	Filling defect diagnosed as innocent tumour	Myosarcoma	Laparotomy. Local removal. Outcome unknown
27 (M.W.)	73	F	1	2.5 cm.	P.M.	Spherical tumour	—	Fibromyoma	Symptomless myoma. Death from coronary occlusion
28 (P.M.)	72	M	1	5 x 5 x 2.5 cm.	Haematemesis and melaena	Lobulated, ovoid with ulceration	—	Spindle cells. Fibromyoma	Three days haematemesis, unresponsive to transfusions. Laparotomy revealed an ulcerating myoma. Local removal
29 (J.R.)	75	M	1	3 cm.	Haematemesis	Sub-mucosal—central ulceration	Suggested carcinoma	Considerable cellular activity. Probably benign	Ba. meal following haematemesis suggested carcinoma. Appearance unaltered 2 years later. Laparotomy. Local excision of myoma. Alive and well 6 months later
30 (M.B.)	60	F	1	7 cm.	Weakness, melaena. Palpable tumour	Sub-mucosal—central ulceration	Rounded filling defect suggestive of innocent tumour	Leiomyoma—benign	Collapse preceded by melaena. Gross anaemia. Tumour removed by partial gastrectomy
31 (E.H.)	65	F	1	5 cm.	Haematemesis	Regarded as carcinoma	An ulcerating lesion	Very cellular and active. Probably benign	Haematemesis. No preceding symptoms. Transthoracic subtotal gastrectomy for supposed carcinoma. Death from shock
32 (O.F.)	61	F	1	4 cm.	Melaena and weakness	Sub-mucosal on anterior wall	Filling defect on greater curvature. Regarded as benign. Supported by gastroscopy	Myoma—benign	Weakness and anaemia following repeated melaenas. Local excision of tumour. Alive and well 14 months later

TABLE 3—(continued)

Case	*Age*	*Sex*	*Number*	*Size (diam.)*	*Method of Presentation*	*Macroscopic Description*	*Radiological Features*	*Histology*	*Clinical Course, Treatment, End Result*
33 (R.M.)	59	F	1	4 cm.	Epigastric pain and vomiting	Sub-mucosal—central ulceration	Infiltration in mid third-regarded as carcinoma	Cellular myoma—occasional mitosis—probably benign	Eight months pain and vomiting. Gross anaemia. Occult blood ++++. Laparotomy. Local excision
34 (L.A.)	45	F	1	"Large"	Haematemesis	Smooth, round, from lesser curvature to posterior surface	Infiltration and ulceration of large area. Regarded as benign	Myoma—benign	Haematemesis seven and three years earlier. Partial gastrectomy. Alive and well 11 years later
35 (D.F.)	37	F	1	10 cm. 180 gm. weight	Epigastric pain. Haematemesis	Sub-mucosal, encapsulated	Infiltration of lesser curvature regarded as carcinoma	Myoma—well differentiated	Haematemesis followed by 6 months dyspepsia. Laparotomy. Enucleation
36 (H.J.)	44	M	1	11 cm.	Haematemesis and melaena	Ulcerating tumour filling fundus	At first N.A.D. Fifteen months later—filling defect at cardia suggesting encephaloid carcinoma	Actively malignant leiomyosarcoma	Haematemesis but Ba. meal N.A.D. Excision of fundus. Oesophagogastrostomy. Patient died 6 months later with massive metastases in liver
37 (E.J.)	53	M	1	7 cm.	Dyspepsia with haematemesis	Intra-mural	Globular filling defect in body of stomach	Leiomyoma—benign	5 years post cibal discomfort, culminating in haematemesis. Local excision. Death from pulmonary embolus
38 (T.H.)	56	M	1	4 mm.	Found during operation for duodenal ulcer	Sub-serosal	—	Leiomyoma. Hyaline and calcified areas	Nodule removed during gastrectomy for duodenal ulcer
39 (A.S.)	59	F	1	8 cm.	Found during operation for ventral hernia	Encapsulated	—	Leiomyoma—benign	Local excision of nodule during herniorrhaphy
40 (R.P.)	45	M	1	6 mm.	Found during operation for peptic ulcer	Sub-serosal	—	Myoma—benign. Degeneration and calcification	Nodule removed during gastrectomy for peptic ulcer
41 (H.B.)	53	M	1	2 cm.	Found during operation for peptic ulcer	Anterior wall	—	Active leiomyoma—a few mitoses. Probably benign	Death from post-operative peritonitis
42 (S.H.)	80	F	1	1 cm.	P.M.	Smooth nodule	—	Leiomyoma benign	Symptomless. Death from carcinoma of stomach
43 (W.M.)	63	M	"A group"	Largest 1 cm.	P.M.	A small group of nodules	—	Leiomyomata—benign	Symptomless. Death from broncho-pneumonia
44 (U.D.)	43	F	1	5 mm.	P.M.	Small nodule	—	Leiomyoma—benign. Hyaline and calcified areas	Symptomless. Death from paralytic ileus after hysterectomy
45 (H.B.)	55	M	1	4 mm.	P.M.	Small nodule	—	Myoma—benign	Symptomless. Death from cerebral thrombosis
46 (E.L.)	52	F	1	4 mm.	P.M.	Small nodule	—	Myoma—benign	Symptomless. Death from intestinal obstruction
47 (K.G.)	76	F	1	2 cm.	P.M.	Nodule growing outwards	—	Leiomyoma—benign	Symptomless. Death from intestinal obstruction
48 (S.L.)	57	M	1	3 mm.	P.M.	Firm white nodule near cardia	—	Leiomyoma—benign	Symptomless. Death from carcinoma oesophagus
49 (E.G.)	79	F	1	5 mm.	P.M.	White nodule at fundus	—	Leiomyoma—benign	Symptomless. Death from chronic nephritis
50 (S.W.)	73	F	1	4 mm.	P.M.	Nodule in wall of pyloric antrum	—	Leiomyoma—benign	Symptomless. Death from carcinoma of the breast

ACKNOWLEDGEMENTS

I am indebted to the staff of the Royal Melbourne Hospital for access to the clinical notes of some of the cases, and to Dr. J. D. Hicks for pathological material. For the remaining cases (Cases 29-50 inclusive) I would thank Dr. A. V. Jackson who has discussed these in a recent number of the Alfred Hospital Clinical Reports.

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GLYCOGEN SYNTHESIS IN HUMAN ENDOMETRIUM

A HISTOCHEMICAL STUDY USING FROZEN-DRIED MATERIAL

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GLYCOGEN has been demonstrated in the endometrium during all stages of the menstrual cycle. It appears diffusely in readily recognizable amounts on about the fourteenth day and marks the onset of the secretory phase of the cycle.

Most cytological investigations on this subject have been carried out on material fixed solely by chemical means. Preparatory freezing and drying by the Altman-Gersh technique provides a superior method of preparing tissue for histochemical investigation.

By this technique, the tissue is snap-frozen in liquid air, subsequently dried *in vacuo* and embedded in paraffin. In this way, diffusion of cell constituents is minimized and smaller concentrations may be more accurately localized.

Materials and Methods

Specimens of endometrium were obtained from 110 cases admitted to the Women's Hospital, Melbourne, for diagnosis. In most instances, the biopsy material was divided into three parts, two of which were placed in Masson's fixative and 80 per cent. alcohol respectively, the remaining portion was "snap-frozen" in liquid air within a few minutes of its removal from the uterus. Some "controls" were kept at room temperature for varying periods up to three hours before "snap-freezing" in order to observe the effects of delay on cytology.

The equipment used for the "freeze-drying" was similar in many respects to the original Packer-Scott apparatus described by Glick (1949) and Forbes (1952). The material in liquid air was transferred to the drying chamber, maintained at a temperature of -35°C. and subjected to a pressure of approximately 10^{-3}mm. of Hg. It was found that the process of dehydration was completed in twenty-four to thirty-six hours according to the thickness of the specimen which was usually about 2 mm. The material was then embedded in paraffin. Sections, which were cut with a rotary microtome set at 7μ, were simultaneously chemically fixed and mounted by floating out on 90 per cent. alcohol. The sections were thus exposed to the chemical fixative for about one minute and it is due to this great shortening of the time of fixation that many of the favourable results of freeze-drying are attributable.

The frozen-dried and Masson fixed sections were then stained with Best's carmine following the procedure described by Perry (1948). Control sections were stained by the Hotchkiss method (Glick, 1949) and corresponding sections treated with saliva for half an hour to verify the identity of the glycogen.

The Advantages of Freeze-Drying

The frozen-dried material consistently provided greater clarity of cell detail than the corresponding chemically fixed preparations. This superiority appeared to be due to several factors many of which have been described by other authors (Gersh, 1932; Bensley and Gersh, 1933; Hoerr, 1936; Simpson, 1941; Glick, 1949).

Shrinkage of tissue and distortion of cell membranes is minimized.

The diffusion of intra-cellular constituents which is inevitable with chemical fixation alone is almost absent.

In ordinary methods, the tissue fluids are slowly replaced by the chemical fixative creating a "diffusion wave" which results in alteration of the sites of cell constituents (De Robertis *et alii*, 1948) and allows time for autolytic changes to occur. The "snap-freezing" involved in the Altman-Gersh technique immobilizes cell constituents and prevents autolysis. The subsequent dehydration enables the chemical fixative to penetrate the tissue very rapidly.

It has been shown that cell constituents are "washed out" in the process of ordinary chemical fixation even though these constituents are not soluble in the fixatives chosen. The rapidity of chemical fixation prior to staining in the technique described above minimizes this process.

Of these factors, the preservation and lack of diffusion of cell constituents provided by this method were of prime importance in this particular study and enabled the early detection and accurate localization of small amounts of glycogen.

Further, the method overcomes the difficulty most workers have had of obtaining sufficient material to employ several fixatives. This technique enables a piece of tissue big enough for only one block to be preserved, so that subsequent individual sections may be treated with appropriate fixatives preparatory to special staining.

Effects of Delay in Snap-freezing

Delay of fifteen minutes or more in placing the biopsy material in liquid air resulted in histological variations. The glycogen in the cell was observed to become fragmented and to lose its precise localization. Extracellular diffusion of glycogen into the lumen of the glands and the surrounding stroma occurred, and when delay was prolonged these changes were more pronounced.

HISTOLOGICAL APPEARANCES

In normal *endometria* glycogen can be regularly demonstrated by ordinary methods at the beginning of the so-called secretory phase which commences at about the fourteenth day.

In sections cut from the frozen-dried material, minute amounts of glycogen are seen in the cytoplasm at the lower poles of the nuclei of the gland epithelium before it can be detected in the ordinary chemically fixed sections (Fig. I) and while the architecture is still proliferative in type.

At this early stage, the glycogen is confined to the nuclear-cytoplasmic interface where it covers the lower pole of the nucleus (Fig. III, B). In sections with only this small amount of glycogen present, the general architecture of the endometrium is that usually recognized as the late proliferative stage.

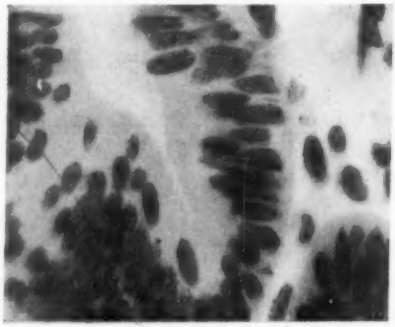

FIG. I. Photomicrograph showing the earliest stage of glycogen synthesis in which minute amounts of glycogen may be seen at the basal poles of the nuclei.
Frozen-dried tissue with Best's Carmine stain. (x 900)

As secretion continues, the glycogen extends to the basement membrane in a thin band. A little later it is seen as a sheath closely investing the nucleus and accumulation begins at the upper pole (Fig. III, B and C).

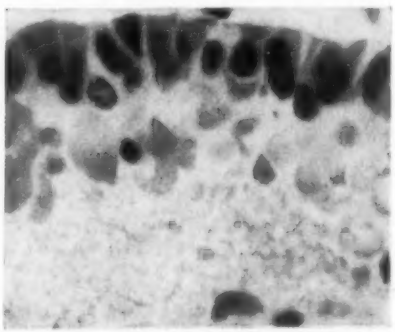

FIG. II. Photomicrograph showing the later stages of glycogen formation and excretion. Glycogen may be seen throughout the cell as well as in the lumen of the gland.
Frozen-dried tissue with Best's Carmine stain. (x 900)

The basal portion of the cell subsequently fills with glycogen and this is followed by accumulation in the supra-nuclear zone (Figs. III, D, E and II).

Parallel with these phenomena certain nuclear changes occur. Before secretion commences the nuclei are elongated (Fig. III, A), but with the appearance of glycogen they become more and more spherical until the glycogen is expelled from the cell. The general configuration of the cells and the site of the nuclei also change. The cells often become shorter and wider as glycogen accumulates and they retain this shape following excretion (Fig. III, H). The nuclei move away from the basement membrane as the glycogen concentration increases. They return to their original position but not to their original shape when the glycogen has been discharged from the cell.

free globules of glycogen are present in the lumen of the glands and the cells appear truncated. This appearance is usually associated with rarefaction in the cytoplasm (Fig. III, E and G) and the phenomenon appears to be repeated until the supra-nuclear portions of the cells contain no more glycogen. After this, gradual disappearance of the basal glycogen is observed (Fig. III, G and H).

Basal glycogen is a constant finding during all stages of secretion. It precedes the appearance of supra-nuclear glycogen and persists after the discharge of the supra-nuclear glycogen from the cell.

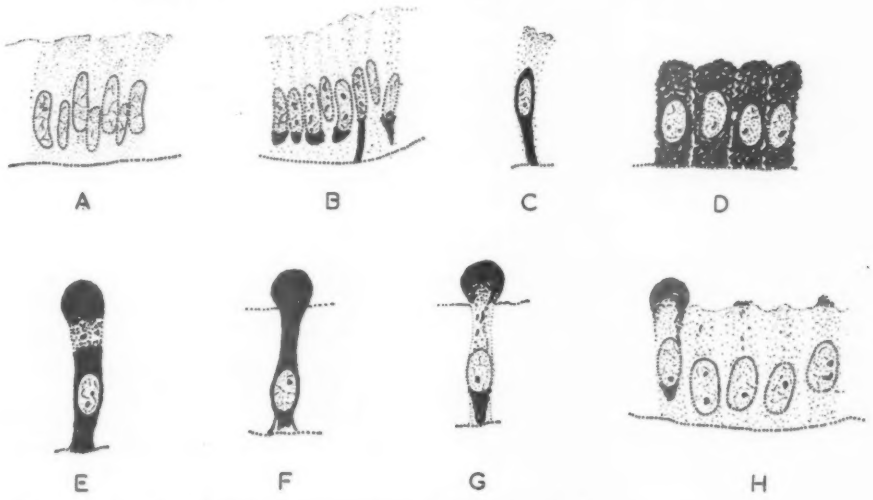

FIG. III. Drawings showing the various stages of glycogen synthesis:—
A. proliferative endometrium prior to the appearance of glycogen;
B. a similar stage to that displayed in Fig. I.;
C. the basal accumulation of glycogen and the peri-nuclear extension of the zone of synthesis;
D. intra-cellular accumulation of glycogen in secretory endometrium;
E., F., G., H. representation of the process of extrusion of glycogen from the cells.

Nucleoli are observed more frequently when glycogen is present and persist in greater numbers following its discharge from the cells.

The liberation of glycogen into the lumen of the glands can be observed in its various stages (Fig. II). First, the glycogen distends the free margin of the cell so that it forms a globule projection into the lumen of the gland (Fig. III, F). In the next stage,

DISCUSSION

The Altman-Gersh method minimizes the alterations in concentration and distribution of intra-cellular constituents which handicap histochemical studies on tissue treated solely by chemical fixatives. Consequently, cell substances visualized by direct staining may be observed in greater detail by this method and conclusions regarding the sites of synthesis of materials such as glycogen appear to be justified.

It is well known that the glycogen appears first in the basal portion of the cell and later in the cycle becomes visible in the supra-nuclear cytoplasm prior to excretion. This succession of events could be explained in three ways. First, by the diffusion of the substance through the body of the cell from the basal cytoplasm where it is synthesized, to an alteration in the site of synthesis or thirdly, to an extension of the sub-nuclear zone of synthesis.

The observations made in this paper suggest that the third process actually occurs.

The appearance of glycogen in the cytoplasm at the surface of the nucleus suggests that the nuclear-cytoplasmic interface is the zone of glycogen synthesis. This process begins at the basal pole and gradually extends over the surface of the nucleus to the upper pole (Fig. III, C).

While the site of synthesis is confined to the lower portion of the nucleus the base of the cell commences to fill with glycogen (Fig. III, B). As this occurs, the zone of synthesis extends and glycogen appears at the upper pole. This is followed by accumulation of large amounts of glycogen in the supra-nuclear cytoplasm.

Although this phenomenon could possibly be explained by a diffusion of glycogen from the base of the cell, the intimate association of glycogen with the nuclear membrane and also the accumulation at the upper pole of the nucleus as distinct from other sites in the supra-nuclear cytoplasm, suggest that the various manifestations of glycogen secretion are caused by an extension of the zone of synthesis of this substance.

Since some glycogen persists in the basal portion of the cell after excretion of the supra-nuclear glycogen (Fig. III, H), it seems feasible to conclude that synthesis persists longer in this region than at the upper pole.

Rossman (1940) observed that the Golgi apparatus undergoes hypertrophy, becomes more complex and congregates in the peri-nuclear zone during glycogen secretion. The appearance of nucleoli in conjunction with the peri-nuclear glycogen and the concomitant changes in the Golgi apparatus in this region gain further significance when considered with the descriptions of cytoplasmic protein synthesis by Caspersson (1942, 1947). In these he ascribes the formation of cytoplasmic protein to a "nucleolus-nuclear membrane mechanism." He observes that this system operates in the interphase of cell activity and "is used not only during growth but also for production of proteins for other purposes such as secretion."

Glycogen makes its appearance when the mitotic or proliferative activity of the endometrium is reduced and the tissue between cell divisions is prolonged.

The appearance of manifestations of protein synthesis, changes in Golgi apparatus and the formation of glycogen, all of which are closely related to the nuclear membrane suggest that this region is of particular significance in the synthesis of cell constituents.

These observations are consistent with Rossman's (1940) postulation that ischaemia develops in the endometrium and brings proliferation to an end. He further states that this ischaemia may explain the appearance of glycogen in the cell and then later the ultimate necrosis of the endometrium.

A study of this kind is clearly in the realms of pure cytology but it does provide an example of the observations possible when material obtained from the body is dealt with immediately on removal.

It seems desirable for the surgeon to have an understanding of the principles involved in such an investigation in order that appropriate material may be obtained for study.

For this reason the results of an investigation which might otherwise be recorded in a purely histological journal are presented here.

CONCLUSIONS

The technique of freeze-drying permits a more accurate appraisal of cell constituents and their functional site.

On the basis of observations by this technique it has been concluded that endometrial glycogen is synthesized at the surface

of the nuclear membrane beginning at the lower pole of the nucleus, later extending to the upper pole.

These conclusions taken in conjunction with the observations by Rossman on the Golgi apparatus and Caspersson on protein synthesis indicate that the peri-nuclear zone is of particular significance as a site for the synthesis of cell constituents.

SUMMARY

1. The freeze-drying technique used in an investigation of the histochemistry of the endometrium is described. The advantages of this technique and the effects of delay in fixation are recorded.

2. Observations are made regarding changes in endometrial cytology during the menstrual cycle.

3. The site of glycogen synthesis and some implications resulting from this are discussed.

ACKNOWLEDGEMENTS

We wish to thank the staff of the Women's Hospital, Melbourne, for their assistance in obtaining the biopsy material on which this paper is based.

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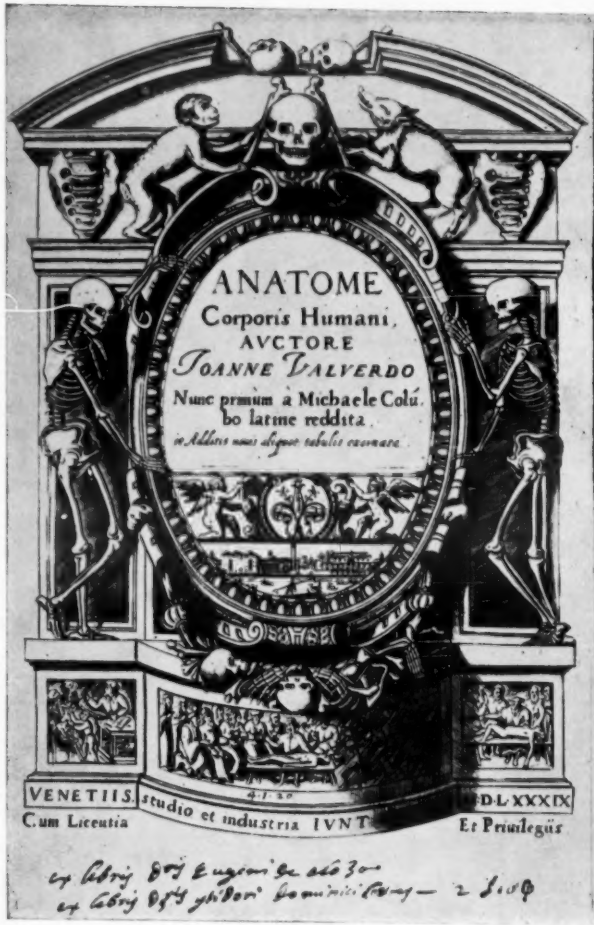
ANATOME
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IOANNE VALVERDO
Nunc primùm à Michaele Colú-
bo latine reddita.
in Addens nunc elegantes tabulas exornata.
VENETIIS
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IOANNES VALVERDVS
HISPANVS.

The College Library

JUAN VALVERDE de HAMUSCO

By K. F. Russell

Department of Anatomy, University of Melbourne

ONE of the most popular anatomical text books of the sixteenth century was written by a Spaniard and published in Italy. Juan Valverde de Hamusco was a student of both Realdo Columbus and Bartolommeo Eustachi and studied at Padua and Rome but beyond these facts we know nothing, even the dates of his birth and death are forgotten.

His book was first printed at Rome in Spanish in 1556 with the title *Historia de la composicion del cuerpo humano, escrita por Ioan de Valuerde* and was frankly a condensed version of the *Fabrica* of Andreas Vesalius both with regard to plates and to text.

Vesalius was justifiably annoyed at this piece of piracy and expressed himself with vehemence in his *Examen* on Fallopius, 1564:—

> Are you, pray, so greatly influenced by the authority of Valverde (who never employed a hand in dissection, who knows nothing of medicine or the primary disciplines, and who made a Spanish translation in connection with this art of ours only for dishonourable profit, and has left our common school) . . . that you dispute with him as if he had actually ever bestowed earnest work on the art? (Cushing: *Bio-Bibliography of Andreas Vesalius*).

In spite of, or most probably because of, his use of the Vesalian text and plates, Valverde's book was very popular and went through the following editions:—

Spanish	Italian	Latin
Rome 1556	Rome 1559	Venice 1589
	Rome 1560	Venice 1607
	Venice 1586	
	Venice 1606	
	Venice 1608	
	Venice 1682	

Ironically enough the plates which Valverde copied from Vesalius were themselves plagiarized by the publisher Christopher Plantin in a book with the title *Vivae Imagines* printed at Antwerp in Latin in 1566 and 1572 and in Dutch in 1568 and at Amsterdam in Dutch in 1647 (the latter printed by Cornelis Danckertz).

The College Library has an excellent copy of the 1589 Latin edition. As in the *Fabrica* the text is divided into seven books and these deal firstly with the skeleton, then the muscles, abdominal and pelvic viscera, thoracic viscera, brain, vascular system and finally the nervous system. There are 46 plates in all, of which 38 are direct copies of the similar plates in the *Fabrica*, but the remaining 8 are more or less original. Five of the latter series are quite original and consist of:

(a) an *écorché* figure holding his skin in one hand and the flaying knife in the other,

and (b) four muscle men, vigorously drawn and quite different from the other plates. (These had first appeared in the 1586 edition.)

As well as the plates, there is a fine engraved title and a portrait. The portrait and a number of the plates were engraved by Nicholas Beatrizet whose monogram (NB) appears on the portrait and on two plates.

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THE PLACE OF MEDULLARY NAILING IN FEMORAL FRACTURES

By N. W. Nisbet

Dunedin

THE management of certain fractures has been simplified by the judicious use of the medullary nail. Seven patients with fractures of the femur are described in this report to illustrate this.

Historical

The method is not new and I am indebted to an excellent symposium in the *Journal of Bone and Joint Surgery* (British Volume, 1950) for the following details. Nicolaysen (1897), Delbet (1906), and Lambotte (1913), used a form of medullary nailing, and Hey Groves (1918) described and illustrated in the *British Journal of Surgery* two femoral fractures splinted with medullary nails of similar pattern to those used to-day. Many of the early cases were complicated by electrolysis between the tissues and the metal—at that time mistaken for infection—but with the invention of inert alloys of suitable malleability and temper the method was re-introduced and practised with safety.

The reason why Küntscher in Germany introduced routine medullary nailing into his hospital was because of the necessity of transferring his patients elsewhere at a moment's notice. He popularized the method, which is now used extensively in many clinics. Whether it will become safe for routine use will be decided when a sufficient number of cases so treated can be assessed.

The chief merits of medullary nailing are that cumbersome and irksome external splints are not needed, joint movement begins early, and the patient's stay in hospital is shortened. Its main demerit is that when complications arise they "offer more possibilities of trouble than any other." These have been well described by various authors in the *Journal of Bone and Joint Surgery* (British Volume, 1950), and a few that occurred in our patients are described below.

Clinical Cases to Indicate the Use of the Nail

The difficulty of controlling the short upper fragment in sub-trochanteric or high shaft fractures of the femur is well known, and this method has undoubtedly solved this problem. Medullary nailing should be used in every case unless there is a definite contra-indication.

FIG. I. Case 1. Comminuted sub-trochanteric fracture of the left femur.

Case 1

H.M., aged 28 years. History: On 1st July, 1951, he fell down a bank 25 feet high and received a sub-trochanteric fracture of the left femur (Fig. I). A medullary nail was inserted by the open method on 6th July, followed by six weeks bed rest and exercises. He was discharged from hospital on 1st August walking with crutches. On 16th May, 1952, the left thigh was clinically normal. There was full power in the thigh and a

full range of knee movement. The patient is to be re-admitted to hospital for removal of the nail (Fig. II).

FIG. II. Case 1. Following reduction and insertion of a medullary nail by the open method.

Comment: The nail was inserted by the "open" method. The fracture was exposed and reduced under vision and the guide wire and nail were guided across the fracture also under direct vision. Too many serious complications have been reported when the nail was inserted by the blind method with X-ray control. No attempt was made to get this patient up too early.

The next case illustrates what may happen with too early walking.

Case 2

L.R., female, aged 29 years. History: Received a simple transverse fracture of the shaft of the right femur at the middle and lower thirds. The fracture was reduced on 22nd July, 1951, and a medullary nail inserted by the open method. The patient was allowed to walk fifteen days after the operation. Progress: One month later the nail bent while she turned with the weight on the leg. This was straightened by manipulation and ten days later she was allowed to walk with a caliper. The thigh gradually bent again, and once more the nail was straightened and a plaster spica was applied and retained for three months. Thereafter a caliper was applied for a further two months and union seemed to be sound. On 16th April, 1952, the thigh suddenly bent while walking along the street. When examined on admission to hospital there was gross bowing of the thigh and X-ray (Fig. III) showed the bent medullary nail and established non-union of the fracture. Operation on 18th April. The nail was straightened by manipulation (Fig. IV) and driven further in. The sclerosed bone ends were freshened and iliac bone chips were packed around the fracture. A plaster spica was applied.

FIG. III. Case 2. Skiagram to show the medullary nail that was bent for the third time. The fracture of the femur is ununited and the bone ends are sclerosed.

Comment: This case raises several interesting problems. Whether all fresh fractures should be treated by this method as a routine I feel has not yet been established. It is probably unwise to allow too early unsupported weightbearing. The quality of the nail only is relied upon, and that used in this case was clearly too soft and malleable. Great care must be taken to ensure that the alloy is of the right temper and the nail is of the correct clover leaf pattern on cross section. In this case it was intended to remove the nail and re-insert a better one, but by no effort on my part could the nail be withdrawn. It had been kinked by the repeated bending and straightening and was gripped by the sclerotic bone. Fracture of the nail has been described.

FIG. IV. Case 2. Following straightening of the nail by manual manipulation and freshening the bone ends. Iliac bone chips were packed around the fracture. Note the kink in the nail.

FIG. V. Case 3. A sub-trochanteric fracture in a woman of 78 years.

The following two patients illustrate that old age may be an indication for medullary nailing.

Case 3

Mrs. A.B., aged 78 years. History: On 19th March, 1950, she fell at home and received a sub-trochanteric fracture of the left femur (Fig. V). On 24th March a medullary nail was inserted by the open method. The nail became impacted in the hard cortex of the upper femoral shaft and 6 inches was left protruding from the wound. This was sawn off with a hacksaw (Fig. VI). Progress: The patient died of uraemia one month later. No post-mortem was obtained.

Comment: The unfortunate outcome in this case was probably due to the heavy hammering that was inadvisedly used in an attempt to drive the nail further on. This caused profound shock and precipitated the uraemia. Hammering only impacts the nail all the tighter so that it cannot be withdrawn. As soon as the nail is felt to impact, it must be withdrawn and a narrower one inserted. A hacksaw is an essential part of the equipment for medullary nailing.

FIG. VI. Case 3. This skiagram shows adequate reduction of the fracture. Although not clearly shown the nail was impacted on the hard cortex of the femur. The protruding end was sawn off.

Case 4

Mrs. A.K., aged 92 years. History: On 2nd May, 1951, she fell in a private hospital and received a sub-trochanteric fracture of the left femur. On admittance to hospital she was in robust health, despite her age. A medullary nail was inserted on 31st July by the open method. An X-ray taken a few days later showed that the fracture had become unreduced, but clinically the thigh remained quite stable and painless. On 16th May the patient was in good health and an inmate of a geriatric hospital. She seems likely to reach her century.

Comment: The avoidance of any form of external splinting in the elderly was clearly a great boon. The X-rays illustrated a fault in technique. The fracture was more comminuted than I had thought and the nail was inserted at the junction of the trochanter and femoral neck. It ought to have been inserted through the tip of the trochanter, a technical detail stressed by Hey Groves (1918).

This patient showed that severe comminution may contra-indicate medullary nailing.

Case 5

A.S., male, aged 18 years. History: On 1st April, 1952, he received a severely comminuted closed fracture of the shaft of the right femur from a motor cycle accident of great violence. Examination showed a large loose fragment protruding on the skin. Open reduction was carried out on 3rd April through a Henry exposure, and a medullary nail was inserted. The large fragment was found to have penetrated the quadriceps, and it was carefully reduced without stripping the periosteum or muscle attachments. As this fragment was gutter shaped, approximation of the main fragments raised it out of its bed. A spica was applied.

Comment: In comminuted fractures of this type it may be necessary to use external, as well as internal splinting. Great care must be taken not to strip the fragments of their muscle attachments and so jeopardise their blood supply. Wiring the fracture before nailing may prevent the elevation of the large fragment.

The following patient illustrated the use of the nail in the treatment of ununited fractures.

Case 6

Miss K., aged 45 years. History: On 9th December, 1950, she received a compound fracture of the lower third of the left femur in a car accident. A few hours later the wound was treated by surgical toilet and the fracture was reduced under vision and skeletal traction and a Thomas splint was applied. The wound healed but the fracture persistently displaced and remained ununited. On 20th July, 1951, a massive onlay tibial graft was applied. Despite close impaction of the fragments, the fracture line opened and the graft fractured. A medullary nail was inserted by the open method on 21st December, 1951, and iliac bone chips were packed around the fracture. No external splinting was used. Bony union occurred, but the rehabilitation was complicated by a left sided hemiplegia.

Comment: Non-union is a definite indication for medullary nailing of fractures. It is usually combined with packing iliac chips around the fracture and external splinting. Too early joint movement may scatter the bone chips. The X-ray showed the nail lying closely under the thin cortex of the femur. In contrast to the dense cortex of the upper femoral shaft, the lower is easily penetrated, especially in blind nailing. Transfixion of the patella and tibia have been described, and also rupture of the popliteal artery.

Pathological fractures lend themselves to medullary nailing.

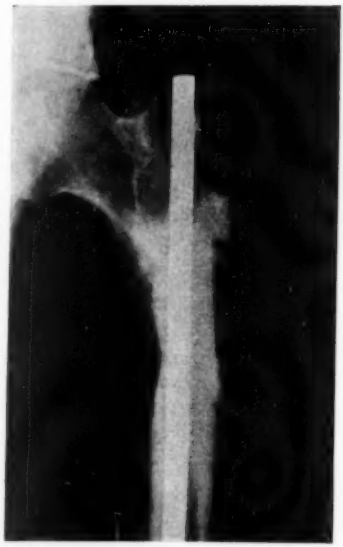

FIG. VII. Case 7. A pathological sub-trochanteric fracture of the femur. Medullary nailing makes the remainder of the patient's life more tolerable by avoiding external splinting.

Case 7

Mrs. A.K., aged 76 years. History: On 2nd April, 1951, she fell at home and received a sub-trochanteric fracture of the left femur. Unsuccessful attempts were made at another hospital to reduce the fracture. Three months later (Fig. VII) the fracture showed its original displacement and was ununited. On 3rd August, 1951, a medullary nail was inserted by the open method. The bone appeared hard and healthy, but because the tissue near the fracture appeared abnormal a specimen was taken for biopsy and showed metastatic carcinoma. Progress: This patient remained comfortable for eight months and has recently died at home. No primary growth was found and no post-mortem was obtained.

Comment: While it is true that the pathological nature of the fracture was not appreciated, medullary nailing made external splinting unnecessary so that the remainder of the patient's life was more tolerable.

CONCLUSIONS

Medullary nailing of femoral shaft fractures, when used with care, can be a great benefit to the patient and solves several problems that have hitherto been a source of worry and trouble to surgeons. Complications can be avoided by anticipation. The case has not yet been made for their place as a routine method of treatment.

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A VARIANT OF THE AURICULO-TEMPORAL SYNDROME

By David L. Dey

Sydney

THE auriculo-temporal syndrome consists of flushing and sweating of the ipsilateral cheek, which occurs when the patient eats, and which follows upon injury or disease of the parotid gland. Although uncommon, this curious phenomenon has been reported by many observers since the first recording of a case by Baillarger (1853).

In almost every instance there has been actual division of tissue by some traumatic agent—most commonly the surgeon's knife as it drains an abscess or removes a tumour of the parotid gland.

Haynes (1897) describes a child aged 19 months in whom there was also congenital hemi-facial hypertrophy and a family history of "nervous instability." The sweating was accompanied by swelling and occurred momentarily on yawning. Otherwise there is little reference to cases in which no division of tissue had occurred, though localized flushing and sweating of the head and neck has been noticed in cases of syringomyelia and other central nervous lesions. The present case is of interest in that there had been no division of tissue, and more than that no trauma or other damage to the parotid gland had been apparent to the parents. The condition has seldom been reported in children. The absence of tissue damage and the "congenital" nature of the phenomenon in this child would appear to have some bearing on the mode of production of symptoms.

Case Report

A female child, aged twenty-two months, was seen in March, 1952. Her mother stated that since she had begun to eat solid food at the age of seven or eight months, a flush had appeared on her right cheek at meal times. This was not present when the child was not eating. It was more apparent when the food was of a "tart" nature, e.g., an apple. The child was very well in other respects.

Local examination showed only normal appearing skin. There was no apparent lesion of the parotid gland or of other deeper tissues. When she chewed actual food, such as a biscuit or an apple, an area of reddening appeared on the right cheek, running in a band about one and a half inches wide from just in front of the ear almost to the corner of the mouth. There was no sweating. Clear saliva could be seen issuing from the opening of the parotid duct. When the stimulus was discontinued, the flush faded rather rapidly. Nothing was seen when she chewed upon a piece of rubber. The child had been a "low forceps" delivery, but there had been no instrumental damage, apart from some slight bruising on the left (contra-lateral) side of the face. The parents positively denied any infection or trauma in the area since that date.

No treatment was advised. The child has been seen since, and the symptoms continue in similar degree.

The Classical Syndrome

Following Baillarger's initial report (1853), description of the syndrome has been elaborated by Parkes Weber (1897) and Madame Lucie Frey (1923) after whom the syndrome is often named. As might be expected, so striking and mysterious a phenomenon has given rise to much argument as to the underlying pathology.

Exposition of these arguments may be found in the papers of Jones (1942), Freeberg, Shaw, and McManus (1948), and Pfeffer and Gillis (1951).

Whilst any form of trauma may be followed by the syndrome, suppurative parotitis requiring drainage is the commonest antecedent. Symptoms have begun from one month to three years after such an incident. In rare instances shorter periods of a few days have been noted.

The affected individual notices flushing and sweating of the side of the face in front of the ear when eating. The degree and the areas affected by these two components may not correspond exactly. Physical examination between meals is largely negative. A scar is usually present. There may be some interference with sensation in the affected area, or a partial facial nerve lesion may be present. The taste of food rather than the act of chewing appears to be the important factor, and apples are frequently mentioned as the most potent stimulus. In cases where there is a parotid fistula symptoms tend to be present only when the fistula is closed.

Spontaneous recovery apparently does not occur. In sufficiently troublesome cases symptoms can be abolished by section or alcohol injection of the auriculo-temporal nerve.

Other Atypical Variants

Two interesting variants on this theme have been noted previously. Ringer, quoted by Parkes Weber (1897), knew of a case in which the condition arose following a period of intense worry, without any parotid injury and persisted subsequently.

Uprus, Gaylor and Carmichael (1934) described a case of localized flushing and sweating on eating in the area of distribution of the anterior cutaneous nerve of the neck. This began about one year after the excision of some lymph nodes through an incision over the middle of the sterno-mastoid muscle. The symptoms were abolished by procaine block of the lingual nerve. The authors scout the possibility of damage to this nerve during the operation, but the association of flushing and sweating with another sensory nerve which also carries secretory fibres to the salivary gland appears significant.

Discussion

Various theories have been advanced to explain the phenomena detailed above. There is confirmation of three observations—

(a) that the syndrome is mediated through the auriculo-temporal nerve for symptoms are abolished if the nerve is blocked,

(b) that the cervical sympathetic plays no part for symptoms persist in spite of adequate sympathetic block,

(c) that there is a local hypersensitivity of the sweat glands to acetylcholine.

As the syndrome persists after destruction or atrophy of all parotid tissue, theories of diffusion of cholinergic substances from the injured gland, or of irritation of abnormally sensitive regenerated fibres by secretory swelling of the gland substance, may be discounted.

Harder to refute, however fanciful it may appear, is the theory of cross innervation following nerve injury and regeneration, with salivary secretory fibres reaching sweat glands and blood vessels. Certainly the normal fibres of supply of these structures are all cholinergic. However, the present case would appear to offer good evidence against such a conception.

In short, the remaining theory, that the syndrome is mediated through normal nerve pathways, which are not usually effective in the intact individual, appears the most attractive. These pathways may become more important following sensitization of the end organs by interference with their more usual nerve supply, or by other factors. Wilson (1936) has suggested such a double innervation of the sweat glands of the face. Cases of gustatory sweating and the case recorded here, where no significant injury has occurred, also indicate the probability that distortion of a normal mechanism is involved.

Summary

1. A case is reported in which the auriculo-temporal syndrome occurred without any evidence of trauma.

2. The typical syndrome is described, and two unusual variants previously recorded are noted.

3. The theories of causation are shortly discussed.

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A RETRO-PERITONEAL CYST DERIVED FROM THE INTERMEDIATE CELL MASS

By Richard Flynn and L. D. Wheeler

Sydney

RETRO-PERITONEAL tumours of any type are comparatively rare and provide common interest to the general surgeon and the urologist. An exact diagnosis before operation is sometimes not possible. Our present interest was stimulated by the following patient's case report.

FIG. I. Intravenous pyelogram showing non-functioning left kidney.

G.T., male, aged 49 years, had first noticed a painless swelling in the left iliac fossa five days before admission. During the previous two years he had been troubled with a pain in the left hip joint and left thigh. The pain was aching in character and intermittent. It would be present for a few days and then he would enjoy a painless period of five or six days. About four weeks before the iliac swelling was felt he had noticed swelling and pitting oedema of the left lower leg. Numbness and tingling in the left leg had increased recently.

On examination the patient appeared to be a healthy male with a swollen left leg which pitted on pressure. On abdominal palpation a rounded elastic swelling approximately 25 cm. in diameter was detected in the left iliac fossa. This mass was also felt on rectal examination. The prostate was normal and the swelling was above and lateral to the prostate. On bimanual examination a large rounded elastic mass which was slightly mobile could be felt. A provisional diagnosis of hydatid cyst, or hydronephrosis in a pelvic kidney was made.

FIG. II. Cystogram showing deformity and displacement of the bladder.

A blood examination showed haemoglobin 15 gm. per cent.; leucocytes 6,600; neutrophils 70 per cent.; lymphocytes 20 per cent.; monocytes 5 per cent.; eosinphils 5 per cent. Casoni intra-dermal test (immediate and delayed) negative. The radiologist reported that a radiograph of the chest was within normal limits, and that of the lumbar spine and pelvis showed no abnormality.

An intravenous urogram showed that the right kidney was functioning well, but that the right ureter was dilated with an apparent obstruction at

about the pelvic brim. The left kidney was functioning poorly and no outline of the left ureter was seen in any film. The bladder in the cystogram was displaced to the right and had a peculiar semilunar shape (Fig. I).

The intravenous urogram was repeated after the injection of 800 cc. of air by the presacral route, and so a combination film of retro-peritoneal pneumography and urography was obtained. This showed an opaque area in the left side of the pelvis. Cystoscopic examination was attempted at this stage, but it was found impossible to pass a cystoscope on account of the extravesical pressure. Therefore, a cystogram was made (Fig. II) and this showed marked displacement of the bladder and also deformity. A barium enema was done to show the relation of the swelling to the large bowel. It was reported on as follows: "The barium enema flowed freely to the caecum but narrowing of the sigmoid (Fig. III) had been caused by a large mass in the left iliac fossa. It rather suggests that the mass is pressing on the bowel but not invading it. No bony lesion seen in the pelvis."

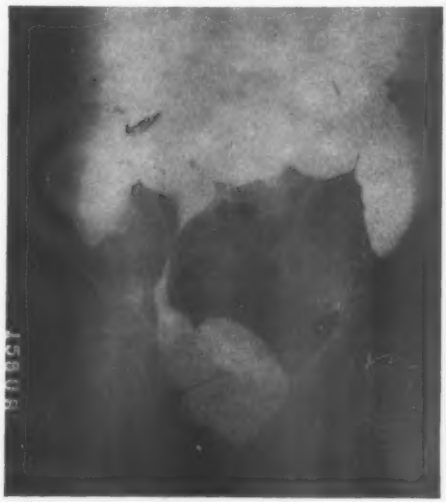

FIG. III. Barium enema showing displacement of the bowel.

It was felt that the outline of the left kidney in the usual position excluded the second as a provisional diagnosis and so on 9th February, 1952, under the presumptive diagnosis of hydatid cyst, operation was undertaken. Through an oblique incision in the left lower abdomen an extra-peritoneal exploration disclosed a retro-peritoneal cyst reddish blue in colour. It resembled a hydatid and so as to prevent soiling the wound with live hydatid scolices a little fluid was aspirated and replaced with a like amount of 10 per cent. formalin. The cyst wall was of a dull whitish colour and about 1 mm. thick. The cyst was separated from the bladder, peritoneum and iliac vessels by blunt gauze dissection. The cyst was not attached to any neighbouring viscus.

FIG. IV. Portion of the cyst wall showing tubules lined by atrophic epithelium and also a malformed glomerulus.
Haematoxylin and eosin. (x 100)

FIG. V. Post-operative intravenous showing return of function to left kidney.

The pathological report on cyst wall was as follows: "The specimen consists of a fragmented cyst wall together with a small amount of atrophic renal tissue forming thickenings in the wall of

the cyst. Microscopically the cyst wall is formed by dense homogeneous connective tissue with no lining epithelium. Sections from some of the thickened portions of the cyst (Fig. IV) show tubules running parellel to each other lined by atrophic epithelium and occasionally a malformed glomerulus is seen. The appearance is such as to suggest the diagnosis of atrophic or agenetic kidney."

The patient's convalescence was uneventful and when he was seen three months later he was quite well. Intravenous urogram at this stage showed that the left kidney was functioning normally and the left ureter was easily identified. The right ureter was no longer showing any dilatation and the bladder had resumed its midline position and its shape was no longer distorted (Fig. V).

In view of the fact that the cyst was not attached to its surrounding tissues by any structure resembling a ureter the diagnosis of supernumerary kidney in its generally accepted sense cannot be made. Further, we think it is an example of primitive urogenital tissue derived from the intermediate cell mass forming a retro-peritoneal cyst.

TORSION OF THE GALL-BLADDER IN A CHILD OF 12 WITH A REVIEW OF THE LITERATURE

By H. D. Rawson

Southampton, England

TORSION of the gall-bladder, a condition of extreme rarity, has been said to occur only in later life, the maximum incidence being between the ages of 60 and 70 (Maingot, 1948).

In this connection, the following case in a child of 12 would seem worthy of mention.

Case Report

Clinical history: On the 28th February, 1952, a boy of 12 was admitted to Odstock Hospital, Salisbury, as an acute abdominal emergency. He stated that he had been quite well until the previous day when, while at school, he felt colicky pains in the right upper abdomen. These pains had persisted intermittently since then and he had vomited on several occasions, bringing up mainly bile-stained material. He had noticed similar milder attacks of pain lasting a few hours at a time and occurring every two weeks or so for as long as he could remember. These were sometimes accompanied by vomiting and he often had to go to bed, though he never told his parents so. There was no history of jaundice or difficulty in micturition. His bowels had opened normally in the morning and apart from anorexia and a slight cough he had no other complaint.

On examination: He appeared a well nourished boy of average build and not unduly distressed. His temperature was 100.0°F., pulse rate 124/min., and respirations 20/min. His tongue was coated but there was no foetor to his breath. The abdomen moved with respiration but there was marked guarding and tenderness in the whole of the right upper quadrant. Elsewhere his abdomen was soft and normal bowel sounds were present. No masses could be felt and there was no tenderness on rectal examination. Nothing abnormal was detected in his heart or lungs. A diagnosis of acute appendicitis associated with an undescended caecum was made and preparations were made for an immediate operation.

At operation: A short upper right paramedian incision was made. The appendix was normal and in the right iliac fossa. A gangrenous gall-bladder was then identified and the incision extended upwards. The gall-bladder was found to be lying quite clear of the liver, its cystic duct being provided with a mesentery, the whole having rotated about the base of this mesentery through one complete turn in a clockwise direction. The gall-bladder wall was black and non-viable. There was a small amount of haemorrhagic exudate in Morison's pouch. Cholecystectomy was carried out with ease after separate identification of duct and artery. The abdomen was closed with drainage after applying "Oxycel" to a small bare area on the liver at the site of the attachment of the cystic duct mesentery. The gall-bladder contained mainly blood. There were no stones.

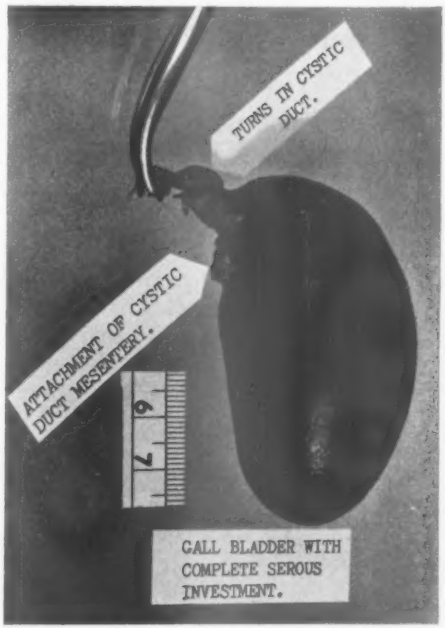

FIG. I. Photograph of the specimen.

Progress: The patient had an uneventful convalescence apart from a minor "post-operative chest." He was discharged on the tenth day. He was quite well when seen as an out-patient a few weeks later.

Pathological report: (Dr. E. M. Martland). "The piece of gall-bladder wall submitted is dense in texture and almost black in colour. Sections show the necrotic changes typical of torsion. The tissues

are suffused with blood and have lost all evidence of structure, the only recognizable objects in the amorphous mass being the shadowy outlines of thrombosed vessels. There is no evidence of inflammatory reaction."

A photograph of the specimen removed is reproduced (Fig. I).

DISCUSSION

Torsion of the gall-bladder was first described by Wendel (1898). Since then the literature on the subject has been reviewed by several observers, the most recent and comprehensive being that of Leger, Debeyre and Mazingarbe (1945) who recorded 86 cases. Twenty-three further cases are listed in the world literature to the end of 1951, bringing the total to 109. I have traced the original articles of 15 of these 23 and Table 1 summarizes the age and sex incidence.

TABLE 1

TORSION OF THE GALL-BLADDER
AGE AND SEX INCIDENCE

Age	No of cases
0 - 10	1
10 - 20	4
20 - 30	3
30 - 40	5
40 - 50	10
50 - 60	14
60 - 70	16
70 - 80	27
80 +	21
Total	101

Youngest — aged 5.

Oldest — aged 92.

Females : Males — 5 : 2 (approximately)

As regards pathology, it is accepted that an essential predisposing factor is a gall-bladder that is freely moveable in relation to the liver. Gross (1936) differentiates two types. In one the gall-bladder has a mesentery; in the other the cystic duct alone is supported by a mesentery allowing the gall-bladder to hang freely. Intermediate forms and combinations of this fold have been described. The case I have recorded conforms with this second type.

By analogy with other congenital abnormalities, e.g., Meckel's diverticulum, one would expect any resulting complications to occur predominantly at an early age. That this is unusual has been taken as an indication that other factors are at work in torsion. Gross (1936) suggests that the loss of fat from surrounding parts and atrophy of the tissues in later age allow for greater ptosis of the viscera, including the floating gall-bladder. Leger, Debeyre and Mazingarbe (1945) point out that the association of gall-stones is a frequent occurrence, a fact which might explain the sex incidence in the more common elderly case. They also indicate that the direction of the torsion is slightly more commonly in a clockwise direction and that one complete turn or less is the usual finding.

That the incidence of torsion is not as rare as published reports might suggest is illustrated by the fact that several surgeons have observed more than one case. For example, Henschen (1915), Krahel (1920), Rendle Short and Paul (1932) and Leger, Debeyre and Mazengarbe (1945) have seen three cases each. Cuervo (1939) has operated on two, both children.

The symptoms on the whole resemble a severe biliary colic with repeated vomiting, but no jaundice. There is commonly a previous history of one or several similar though less severe attacks. The temperature is often slightly elevated and the pulse rate rapid. There is rigidity and marked tenderness in the right hypochondrium and a mass may be felt. Brocq (1931) describes the mass as being moveable transversely, this being doubtless due to the loose attachment of the gall-bladder to the liver.

Of course, the condition is seldom diagnosed pre-operatively. Krahel (1920), however, made a correct diagnosis in one of his three cases. It has been suspected on more than 20 occasions before operation, especially when a mass has been palpable, a sign present in over half the cases Gross (1936).

Kettner (1930) and Anschütz (1947) stress the value of X-ray examination in diagnosis in cases where opaque gallstones are present.

The treatment is, of course, immediate cholecystectomy. In most cases the gall-bladder is gangrenous but perforation is uncommon. Leger, Debeyre and Mazingarbe (1945) mention the relatively good prognosis of 72 per cent. in 69 cases treated surgically.

I have traced the original reports of the four other cases occurring in children under the age of 15. The clinical picture conformed, in general, with the case I have reported. Cuervo (1939) has the distinction of having had the two youngest recorded cases. His first, a boy of five, survived, but his second, a boy of 11, died following intervention after seventy-two hours. Daux (1925) operated on a girl of 11 but could find no abnormality. A gangrenous gall-bladder was found at post-mortem. Dabadie (1946) successfully operated on a boy of thirteen. In this boy, and in Cuervo's first patient, a mass was palpable before operation. Dabadie had the possibility of torsion in mind before operation, but, as also did Cuervo and Daux, he thought a high appendicitis was the most likely diagnosis.

SUMMARY

1. A case of torsion of the gall-bladder in a child of twelve is reported. This is the second youngest surviving case that I have been able to trace in the world literature and is the youngest case described in the English language.

2. The literature to the end of 1951 has been reviewed.

ACKNOWLEDGEMENTS

I am indebted to Mr. Alexander Duff for permission to treat and publish this case and to Mr. J. Clarke of Odstock Hospital for his excellent photograph. The specimen is on view at the Museum of the Royal College of Surgeons, Edinburgh.

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Books Reviewed.

SURGERY FOR STUDENTS OF NURSING.

By JOHN CAIRNEY, D.Sc., M.D., F.R.A.C.S. Christchurch, New Zealand: N. M. Peryer Ltd., 1952. 8¾" x 5½", 326 pp., 120 illustrations. Price: 64s. 6d.

This volume, which provides a wide range of instruction in both general and special fields of surgery, should prove most useful to nursing trainees. It is written in concise and lucid language, and is provided with simple and clear illustrations.

In the teaching of nurses a middle course must be steered between presenting on the one hand a meagre picture which does not provide an adequate background of knowledge, and on the other hand supplying a mass of information which, for the most part, will be useless. The author, who has had a long experience in the teaching of surgery to student nurses, overcomes this difficulty with great success.

The inclusion of a chapter on eye, ear, nose and throat should be of value in training schools where lectures on these subjects cannot be arranged; but some further space could have been devoted to such important considerations as the treatment of head injuries.

In the section on intravenous infusion a useful addition would have been detailed instruction in the care of the running infusion apparatus. In discussing the treatment of paralytic ileus and peritonitis mention might well have been made of the important nursing duty of allowing the patient nothing by mouth; and in the list of causes of gangrene, the tight plaster cast could have been included.

Despite these minor criticisms this book is generally comprehensive and well presented, and should fill a definite need for students of nursing.

GEORGE BASS, 1770-1803.

By KEITH MACRAE BOWDEN, M.B., M.S. Melbourne: Oxford University Press, 1952. 8½" x 5½", xi plus 171 pp., frontispiece and 12 plates. Price: 21s.

So far as Australia is concerned, one of the outstanding truants of medicine is George Bass, sometime surgeon to the Royal Navy, navigator, explorer and, if one may use the term, gentleman adventurer. It is a matter of considerable satisfaction that his full biography should now be written for the first time by Keith Bowden of Melbourne, and it is also a matter of congratulation to the author that this biography should be awarded first prize in the Victorian State Government's Centenary Literary and Historical Competition. This is only appropriate for it was the coast of Victoria that Bass explored in a whale boat in 1798, when he discovered Westernport Bay and the Strait that now bears his name.

The author has done a very considerable amount of detailed historical research and the book that has resulted from his work redounds to his credit and must remain the definitive biography of Bass for many years to come. Although Bass obtained his diploma of Member of the Company of Surgeons on 2nd April, 1789, and qualified as a surgeon's mate in the Navy on 4th June of the same year it very soon became obvious that his real love was exploration and discovery and, of course, it was in these fields that he became most famous. His tragic disappearance in 1803 when in command of the *Venus* will probably always remain a mystery although Bowden gives a very logical suggestion of his fate.

His short life, for he was only 32 when he disappeared, is admirably dealt with in this book and his voyages of discovery make fascinating reading.

Bass is a man the medical profession could well honour even though his exploits were far removed from the practice of medicine; the part that he played in the discovery of this country should never be forgotten.

THE SPREAD OF TUMOURS IN THE HUMAN BODY.

By R. A. WILLIS, M.D., D.Sc.(Melb.), F.R.C.P.(Lond.). Second Edition. London: Butterworth and Co. Ltd., 1952. 9¼" x 6¼", x plus 476 pp., 85 illustrations. Price: 94s.

All those who are interested in the study of tumours and particularly those who have followed the writings of R. A. Willis will be pleased to see this second edition of his first work. It is unnecessary to remind readers of the nature of the reception of the first edition, nor of the unceasing demand that there has been for this. It is no exaggeration to say that this book, published in 1934, still remains the outstanding contribution to this particular subject in the English language.

The second edition necessarily is a reproduction of the first with appropriate additions. Like its forerunner, it carries all the hallmarks of the meticulous and careful observation which is so characteristic of Willis's work; it contains the clear and definite statements (which we may appreciate, even though we do not always approve) found in his other works and it contains the judicious mixture of material culled from the literature and his own personal observation to which we have become accustomed.

Much of the text has been, to some extent, rewritten and re-arranged and there are considerable additions embodying the advances of the last eighteen years. Doubtless many readers, like the reviewer, will find paragraphs or even parts of chapters which are not to his mind as complete as they should be. But to labour these points or even to give them more than just a passing mention would be unrealistic in an age where there is such a spate of literature, in every specialty, that even the specialist is unable to cope with every detail.

The presentation of all the details which might have been included in this volume would, of course, so overshadow as to possibly smother the general principles which are so important and which shine so clearly from Willis's work.

The general form of the volume is in conformity with his *Pathology of Tumours*. It is excellently produced on good art paper and the illustrations, which are somewhat reduced in number but at the same time contain some new subjects, are on special art paper at the back of the volume. These illustrations are very well produced and show clearly the various features which are discussed.

This volume will need no commendation for the student of oncology, who has even a nodding acquaintance with the earlier edition, but we would recommend it strongly to the attention of younger students who may possibly be unacquainted with the earlier work.

CLINICAL PROBLEMS OF WAR.

By ALLAN S. WALKER, M.D., Ch.M., F.R.A.C.P. Canberra: Australian War Memorial, 1952. 9¼" x 6", xxiv plus 726 pp., 73 illustrations. Price: 35s.

This volume is one of a series of official war histories dealing with various aspects of the Army, Navy and Air Forces during the war of 1939-1945. It is the first of the Medical contributions (Series 5).

It covers the activities in a very wide field from Palestine to North Queensland, including Malaya and Siam. Those who were in these areas will recognize the authentic touch, so that it is clear that Dr. Walker has wisely obtained considerable assistance from various officers in the particular areas.

It is a matter of congratulation to him and satisfaction to the reader that he has covered such a large field in so convincing a manner.

The first part covers the medical field and, of this, the first section deals with infectious diseases and justifiably comprises more than a third of the volume. In each part a detailed statement of methods of prevention takes precedence over all other aspects. In many instances the story of the gradual control of disease, even though well known to the reviewer, provides a fascinating story which clearly can be heard again with advantage.

Section two contains a review of the various general conditions such as cardiovascular disease, disease of the urinary tract, etc., and, amongst these, probably malnutrition and the effects of heat are the most important.

Part two deals with surgical problems. Although these did not have the over-riding importance that they showed in the 1914-1918 war, nevertheless they indicated the advantage that had been taken of progress between the wars and thus specialities received particular emphasis. Much of the treatment was based on principles laid down in the 1914-1918 war since, for example, antibiotics became generally available in adequate quantities only in the closing stages of the Pacific War. Emphasis on these principles would appear to be more than justified by observations made in the post-war years.

Part three deals with special subjects such as dermatology, psychiatry and medical rehabilitation.

The volume is well printed and bound and the illustrations are well produced on good art paper. One would like to see the illustrations and diagrams numbered and referred to more adequately in the text. The index is anything but adequate. However, despite these shortcomings, this book will be found of tremendous value to those wishing to obtain some information regarding the medical services in the last war and will be read with great interest, profit and pleasure by those who took part in it.

PICTORIAL HANDBOOK OF FRACTURE TREATMENT.

By EDWARD L. COMPERE, M.D., F.A.C.S., and SAM W. BANKS, M.D., F.A.C.S. Third Edition. Chicago, U.S.A.: The Year Book Publishers, Inc., 1952. 8¼" x 5½", 424 pp., 223 illustrations. Price: $6.50.

This new edition will be an asset to the library of anybody treating fractures. In most instances the unnecessary detail is eliminated and the pictorial descriptions are vivid enough to follow the essential steps. The chapters on treatment of fractures of the neck of the femur, fractures around the elbow joint in children and the wrist scaphoid are carrying good diagrams and a full explanatory text.

A third author, Dr. C. Compere, has taken a hand in this volume, and it is interesting to note how each succeeding edition keeps pace with modern treatment. This book is recommended to anyone whose practice involves traumatic work.

CARDIOVASCULAR RESEARCH: British Medical Bulletin.

London: Medical Department—The British Council, 1952. 11¼" x 8¾", iii plus 118 pp., 8 plates. Price: 21/- net.

This volume of the British Medical Bulletin contains a series of papers by distinguished British cardiologists. They outline some of the more important advances in the understanding and treatment of cardiovascular diseases which are taking place in Great Britain.

In the introduction it is pointed out that such advances are dependent upon the availability and applicability of laboratory techniques to clinical problems. For example, because of progress in the art of cardiac surgery laboratory techniques such as the recording of intra-cardiac pressures and of cardiac sounds have assumed an essential place in the clinical diagnosis of many cardiac conditions. However, these authors are conservative, and in discussions of laboratory techniques they have included only those of established value in clinical medicine.

The articles cover only a limited section of cardiology, and most of them are concerned with the causation, diagnosis and treatment of systemic and pulmonary hypertension and of occlusive arterial disease. Attention is drawn to the functional unity of the vascular system and to the fact "that the cardiologist neglects the peripheral circulation at his peril." Prevention of cardiac disease has not been neglected and a study of the aetiological factors in rheumatic heart disease is included.

Any practitioner wishing to know the trends of advancing knowledge in Britain's leading cardiological centres will find this volume a handy and stimulating guide, which covers aspects of the physiology, pathology, clinical medicine and surgery of cardiovascular diseases.

OBSERVATIONS ON THE GENERAL EFFECTS OF INJURY IN MAN, WITH SPECIAL REFERENCE TO WOUND SHOCK.

By R. T. GRANT and E. B. REEVE. London: His Majesty's Stationery Office, 1951. 9¾" x 6", xiii plus 313 pp., 18 illustrations. Price: 8s. 6d. net.

This volume is yet another excellent product of the Clinical Research Unit of the Medical Research Council.

It is essentially the recording of an investigation into the systemic effects encountered in the variety of conditions included under the heading of "shock."

Over 300 patients have been studied in detail and the findings have been co-related to show some of the differing syndromes encountered. The results of special investigations have been evaluated and put into a working perspective.

In their discussion and conclusions, the authors have gone a long way towards sorting out much of the confusion which has grown up around the term "shock." They have shown that "shock" has been used to describe many different and distinct syndromes, each with individual circulatory patterns. In this way they have performed a great service.

Throughout the whole book there are many sound common sense clinical observations and recommendations as to the treatment of the various syndromes which make interesting and instructive reading for those who have to deal with "shocked" patients.

FLUID THERAPY. A Clinical Manual.

By CARL A. MOYER, M.D. Chicago, U.S.A.: The Year Book Publishers, Inc., 1952. 7⅓" x 4¼", 191 pp., 20 tables. Price: $3.75.

As the sub-title indicates, the author has limited theoretical considerations to a minimum. The introductory chapters refer to history and basic principles. These are both interesting and lucid. In the "Evaluation of Diagnostic Signs" one feels too much weight is placed on single entities, but the whole chapter is a salutory reminder that signs and symptoms can not be well evaluated either singly or lightly.

The author is perpetuating the error of referring to sodium and chloride ions as "sodium chloride." This is particularly unfortunate in the interpretation of the Fantus Test where it leads to incorrect thinking.

The sections on alkalosis and acidosis are clear and straightforward, but one feels the one on potassium could have been enlarged with benefit. The basic principles of treatment and complications are well covered in a simple fashion and contain many helpful hints.

It is good to see small books of this type appearing because fluid therapy is only a small but very important part of both Medicine and Surgery, and generally all too little is known of the subject by practitioners.

SURGERY FOR NURSES.

By JAMES MORONEY, M.B., Ch.B., F.R.C.S., L.R.C.P. (London). Foreword by Miss DOROTHY M. SMITH, O.B.E. Second Edition. London: E. and S. Livingstone Ltd., 1952. 8¾" x 5¾", xii plus 676 pp., 585 illustrations. Price: 27s. 6d. net.

"Surgery for Nurses," by James Moroney, is a book that fills a very real need. With the ever-growing complexities of modern surgery it becomes increasingly difficult for the nurse to understand what the surgeon is doing and what her part is in the general scheme of treatment.

This book is a very successful attempt to explain the principles and practice of surgery in language that student nurses can understand. Its use, however, is not likely to be limited to the trainee; the experienced sister wishing to keep up to date will also find everything she needs in it. As its name implies—the book is a text book of surgery; it is not a text book on nursing surgical patients, and while there is a discussion on nursing care in every section, these discussions are not very detailed.

The book is beautifully produced and profusely illustrated with excellent photographs. Its length (over 600 pages) makes it more useful as a reference book than as a text to be read from end to end by the student nurse, but as a reference book it can be whole-heartedly recommended.

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